The Journal of Bone and Joint Surgery

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The Australian Orthopaedic Association
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LATE RESULTS IN LEGG-PERTHES DISEASE *†

BY EUGENE R. MINDELL, M.D., AND MARY S. SHERMAN, M.D., CHICAGO, ILLINOIS

From the Department of Surgery, Division of Orthopaedic Surgery, University of Chicago

Ever since Legg-Perthes disease was recognized as a distinct entity, its treatment has been the center of considerable controversy [7, 11, 12]. Most early writers were convinced that treatment had no effect on the final result [8, 13, 18, 19]. However, following the papers of Eyre-Brook, Danforth, and others, patients were treated by immobilization or bed rest for as long as three or four years[6]. In contrast, various types of surgery have been recommended as a means to shorten convalescence [4, 14, 17, 20]. Methods currently in use vary from prolonged immobilization of the involved hip to the use of braces or crutches [9, 15, 16]. The present study was undertaken in an attempt to evaluate treatment in terms of end results.

Only those cases are evaluated in which the diagnosis is unquestioned, the type of treatment known, and the clinical and roentgenographic findings available after the disease is over. Seventy-two patients with seventy-eight involved hips meet these requirements and form the basis for this study. Half of them have been followed for six to fifteen years; six individuals have been followed for over twenty-five years (Table I). In two thirds of the cases, growth from the upper end of the femur has been completed.

The type of therapy is classified as ambulatory or non-ambulatory. In some cases both types were used. However, during the prolonged period of active breakdown and

FIG. 1-A

* This study was supported in part by a grant from the Douglas Smith Foundation.

† Read at the Annual Meeting of The American Academy of Orthopaedic Surgeons, New York, N. Y., February 14, 1950.

TABLE I

LENGTH OF FOLLOW-UP IN SEVENTY-EIGHT CASES

Period of Follow-up (*Years*)	No. of Cases
1 to 5	15
6 to 15	41
16 to 25	16
26 to 40	6
Total	78

FIG. 1-B

FIG. 1-C

A poor result in an untreated patient.

Fig. 1-A: August 8, 1940. Roentgenogram made when the patient was first seen, at the age of fourteen.

Figs. 1-B and 1-C: July 1, 1949. Nine years later, when patient was twenty-three.

replacement of the necrotic femoral head, one method of treatment always predominated. In a few cases the choice of classification was difficult, but the treatment considered most significant was selected. Ambulatory treatment refers to the use of crutches without weight-bearing, or, in some cases, to an ischial weight-bearing brace. Non-ambulatory treatment includes prolonged periods of bed rest, traction, or immobilization. The length of treatment in these two groups was essentially the same. Treatment started when the necrotic femoral head was still fairly

FIG. 2-A

FIG. 2-B FIG. 2-C

FIG. 2-D

A satisfactory result in a patient treated by bed rest.

Fig. 2-A: March 2, 1938. View of both hips when the patient was first seen, at the age of five.
Fig. 2-B: June 28, 1939. Fig. 2-C: June 8, 1940.
Fig. 2-D: June 2, 1949, when the patient was sixteen.

intact, with little or no deformity and no significant change in the femoral neck, was considered early. Treatment started when changes of the head and neck were already extensive was considered late. Patients whose treatment was begun when no further changes were expected were classed with those who were untreated.

FIG. 3-A

FIG. 3-B

FIG. 3-C

A satisfactory result obtained by the use of crutches.

Fig. 3-A: June 30, 1936. Roentgenogram made when the patient was first seen, at the age of seven.
Fig. 3-B: December 14, 1937. Fig. 3-C: August 30, 1938.
Fig. 3-D: September 22, 1949, when the patient was twenty.

Figs. 4-A, 4-B, 4-C and 4-D illustrate the extent to which the upper portion of the femur can be remodeled in younger children.

Fig. 4-A: October 30, 1944. Appearance when the patient was first seen, at the age of five.
Fig. 4-B: February 23, 1945. Fig. 4-C: March 20, 1946.
Fig. 4-D: May 18, 1949, when the patient was ten.

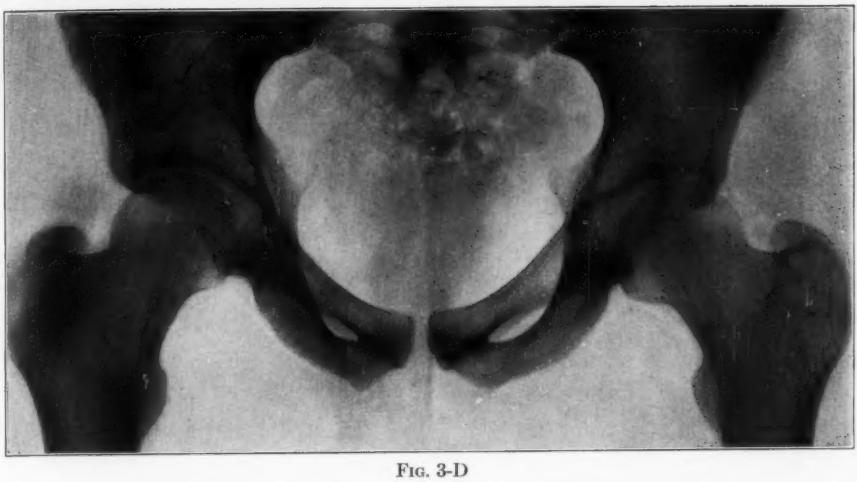

FIG. 3-D

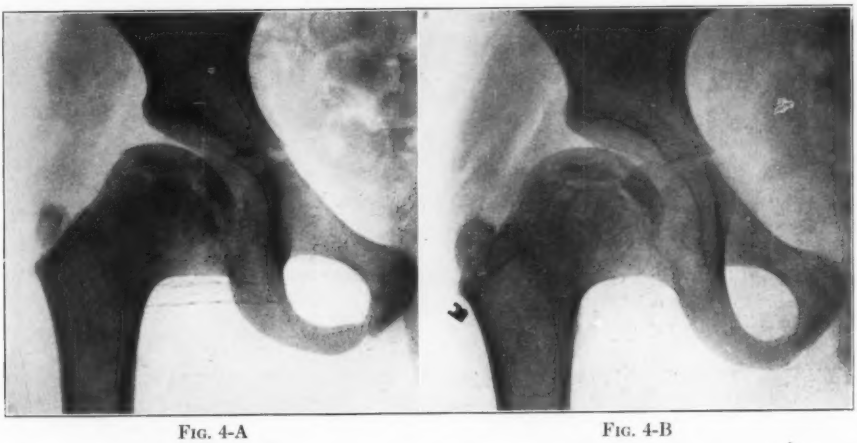

FIG. 4-A

FIG. 4-B

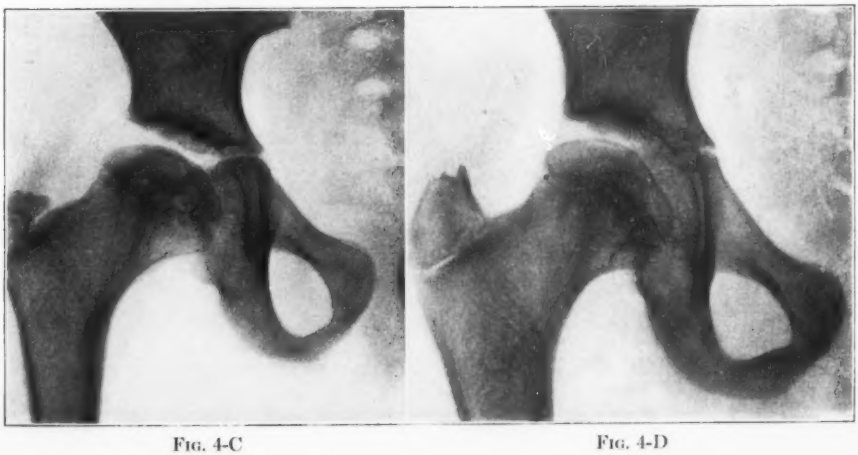

FIG. 4-C

FIG. 4-D

Comparison of the present physical and roentgenographic findings with those of the uninvolved hip reveals that most patients have some residual effects of the disease. However, even those with the worst results are not severely handicapped. For purposes of analysis, the results have been strictly classified as satisfactory, fair, or poor, even though the differences in many cases are not great.

1. Patients considered to have a *satisfactory* result are symptom-free, but on careful examination a few degrees of limitation of rotation or abduction and slight muscular atrophy are usually found. Roentgenographic examination shows the hip to be without deformity and the articular cortices to be free of irregularities.

2. Those patients whose final results are considered *fair* have minor complaints, such as pain with changes in the weather or after long walks. One or two centimeters of shorten-

FIG. 5-A

FIG. 5-B

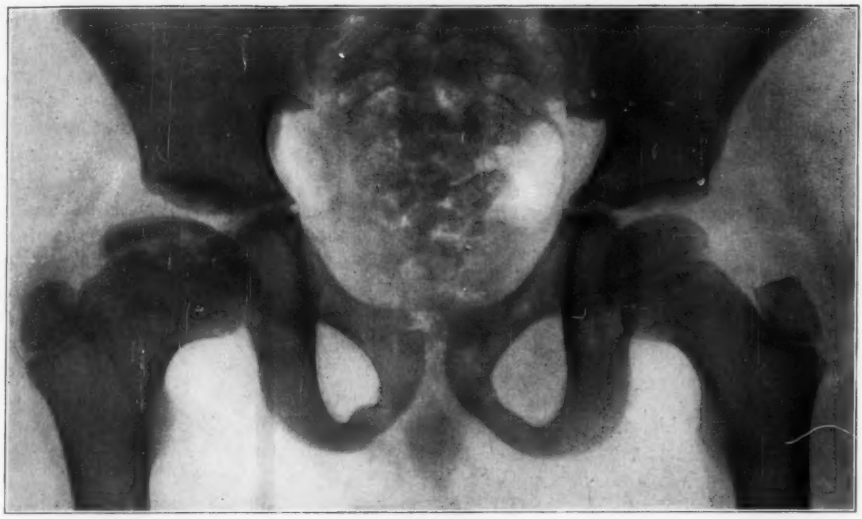

FIG. 5-C

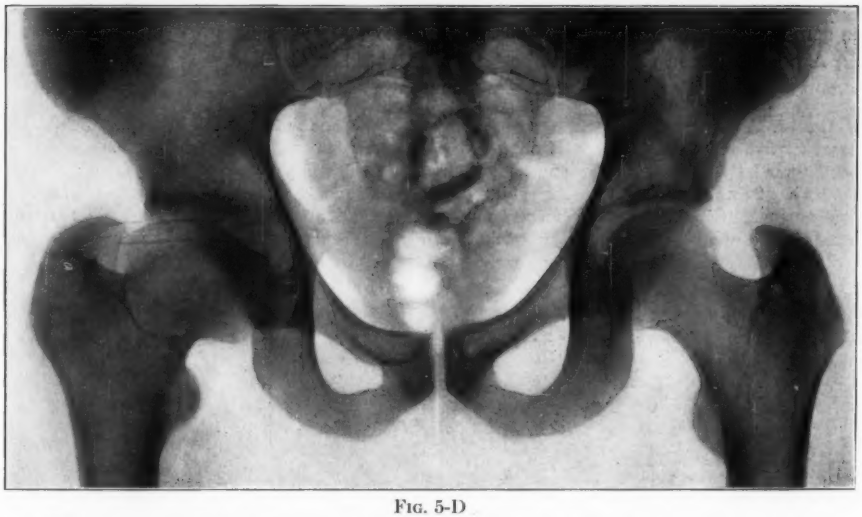

FIG. 5-D

Illustrating the correlation between severe femoral-neck disturbance and a poor final result.

Fig. 5-A: May 22, 1934. Patient was first seen at the age of eight.
Fig. 5-B: April 2, 1935. Fig. 5-C: August 3, 1936.
Figs. 5-D and 5-E: June 20, 1949. Fifteen years after first examination, when the patient was twenty-three.

ing of the involved limb and mild atrophy of the gluteal, thigh, and calf muscles are found. Roentgenograms show a moderately flattened and widened femoral head, slightly irregular cortices of the femoral head, with or without corresponding acetabular changes, and a widened, shortened femoral neck, with or without angulation.

3. Those patients with *poor* final results have more pain and limp and some disability. A few have enough trouble to seek medical care and even to lose time from work. They have as much as 4 centimeters of shortening and considerable muscular atrophy, with sufficient instability to produce a limp. Significant limitation of rotation and abduction

FIG. 5-E

is found in all of this group. In a few patients rotation is practically absent, and limitation of abduction and flexion is severe. Roentgenograms demonstrate gross deformity of the femoral head and neck and occasionally of the acetabulum. Lipping, sclerosis, loose bodies, unreplaced and ununited fragments, or exostoses are sometimes present.

The worst results were seen among the thirteen patients who were untreated, only three of whom have a satisfactory outcome. It is evident that those patients who were treated early have a much higher incidence of good results than those who were treated late or not at all. No significant difference can be demonstrated in the results following the use of ambulatory or non-ambulatory methods of treatment. Both methods produced a high percentage of good results (Table II—page 21). Among the patients treated early by ambulatory methods, no differences are found between those who used crutches and those treated by braces. The resultant slight shortening, atrophy, and limitation of motion are the same in these two groups.

A poor result in an untreated case is illustrated in Case 1. No patient whose treatment was effective early has had such a poor result.

CASE 1. A fourteen-year-old male was seen because of pain and limp in his right thigh, which had been present for five years and had become worse in the preceding few weeks. These symptoms occurred only when he was fatigued or was very active. All motions of his right hip were moderately limited because of pain. He had 2 centimeters of shortening of his right lower limb and 3 centimeters of atrophy of the right calf. Roentgenograms (Fig. 1-A) revealed marked deformity of the right femoral head and neck and premature closure of the capital femoral epiphysis from old Legg-Perthes disease. Treatment was not instituted because the disease was inactive. Roentgenograms made when the patient was twenty-three years of age (Figs. 1-B

FIG. 6-A

and 1-C) demonstrated overgrowth of the femoral head, asymmetrical shortening of the femoral neck, high trochanters, flaring, and some irregularity of the articular cortex of the acetabulum.

The patient continues to have a persistent mild limp and occasional pain when he is active. He has 2.5 centimeters of shortening, a positive Trendelenburg sign, 2 centimeters of thigh atrophy, and moderate limitation of motion of the right hip.

Satisfactory results may be obtained from either ambulatory or non-ambulatory treatment. Case 2 concerns a patient whose treatment consisted of bed rest for one and one-half years, followed by a brace for fifteen months. At the age of sixteen he had no symptoms or abnormal physical findings. The roentgenographic result (Fig. 2-D) was satisfactory. In contrast, Case 3 presents a patient whose treatment consisted of non-weight-bearing on crutches for twenty-six months, and whose result was equally satisfactory.

FIG. 6-B

FIG. 6-C

CASE 2. A five-year-old boy was seen because of a limp and mild pain in his left thigh. The roentgenogram (Fig. 2-A) showed the left femoral head to be irregularly dense. The cartilage space was widened and some irregularity was present in the metaphysis. A diagnosis of fairly early Legg-Perthes disease was made and the patient was put to bed for one and one-half years. He was then allowed up in a walking caliper brace for fifteen months, and thereafter was on crutches, bearing some weight on his left lower extremity, for another six months. A roentgenogram made when bed rest had been discontinued (Fig. 2-B) showed that the necrotic head had been fairly well replaced. The roentgenogram made when the brace was discontinued (Fig. 2-C) demonstrated that the contours of the hip had been reconstituted. At the age of sixteen the patient was asymptomatic and roentgenograms (Fig. 2-D) revealed a normal appearing hip.

CASE 3. This boy was first seen at the age of seven because he had had a mild limp for five months. Mild limitation of motion of the left hip was present. Roentgenograms (Fig. 3-A) showed the femoral head to be dense and somewhat flattened. The cartilage space of the left hip joint was wider than on the right, and slight irregularity was present in the left metaphysis. A diagnosis of early Legg-Perthes disease of the left hip was made and the patient was treated by non-weight-bearing on crutches for twenty-six months. Roent-

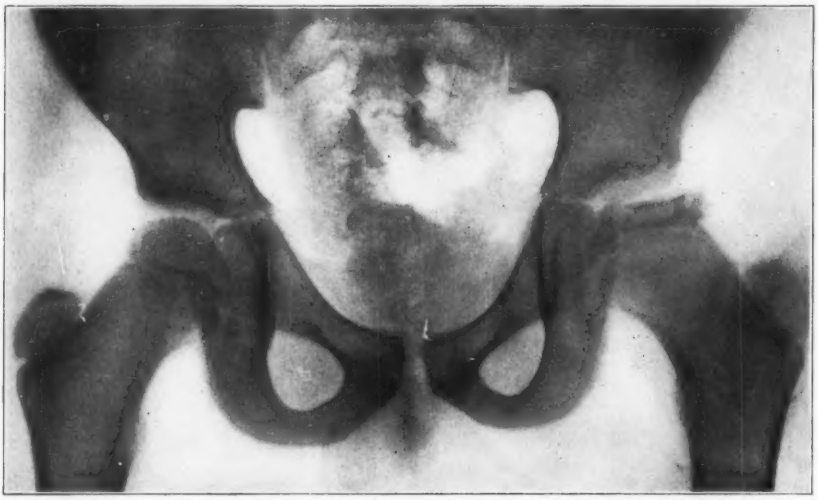

FIG. 6-D

FIG. 6-E

FIG. 6-F

A case in which asymmetrical growth retardation produced deformity of the femoral neck.

Fig. 6-A: December 19, 1936. The patient was first seen at the age of eight.
Fig. 6-B: April 6, 1937.
Fig. 6-C: July 6, 1937.
Fig. 6-D: November 19, 1937.
Fig. 6-E: October 11, 1940.
Figs. 6-F and 6-G: 1949. Thirteen years after treatment was begun, when the patient was twenty-one.

FIG. 6-G

genograms, one and one-half years (Fig. 3-B) and two years (Fig. 3-C) after treatment had been begun, demonstrated that the necrotic femoral head was replaced with little resultant deformity.

At the age of twenty, he does heavy farm work yet has no symptoms referable to his hip. Roentgenograms (Fig. 3-D) demonstrate a normal appearing hip.

Møller, Slocum, and others have stated that the younger the patient, the better the prognosis [1]. Analysis of sixty-two treated patients confirms these observations (Table III).

Ten of eleven patients whose disease began before the age of five have satisfactory results. The one poor result in this group is that of an obese child, whose disease was severe and whose treatment consisted of poorly fitted braces. In contrast, of eleven patients whose disease began after the age of eight, only two have satisfactory results.

The fact that younger patients have better results has several possible explanations. The femoral capital ossification center is smaller and is protected by a relatively thicker layer of cartilage, and in addition, less weight is placed on the necrotic femoral head by a smaller child. Younger children also have a longer active growing period during which the upper portion of the femur may be remodeled, as illustrated in Case 4.

CASE 4. Initial roentgenograms (Fig. 4-A) of a five-year-old girl disclosed deformity of the femoral head and neck due to moderately advanced Legg-Perthes disease. Subsequent roentgenograms (Figs. 4-B, 4-C, and 4-D) demonstrated the remodeling which occurred in the succeeding five years. The roentgenogram made at the age of ten (Fig. 4-D) showed a femoral head and neck of practically normal shape.

As might be expected, the patient is asymptomatic, and examination shows her hip to be normal.

It was possible to follow abnormalities of the femoral neck in fifty-three cases. The usual change consisted of a diffuse, irregular zone of decreased density in the metaphysis,

which sometimes appeared early and sometimes late. This change was often opposite an area of decreased density in the head, and filled in as the head healed. Sometimes a lesion appeared just below the epiphyseal line, filled in as the epiphysis grew away from it, and finally was obliterated. Occasionally successive lesions developed in the subepiphyseal area and each followed the same pattern. Eight patients, or 15 per cent. of this group, had large discrete lesions, often surrounded by thin sclerotic walls and sometimes associated with collapse of the overlying femoral head. Of these eight patients, six had poor final results, one a fair result, and one a satisfactory result. It seems, therefore, that extensive changes in the femoral neck generally indicate a poor prognosis. An example of the correlation between severe disturbances of the femoral neck and a poor end result is seen in Case 5.

FIG. 7-A

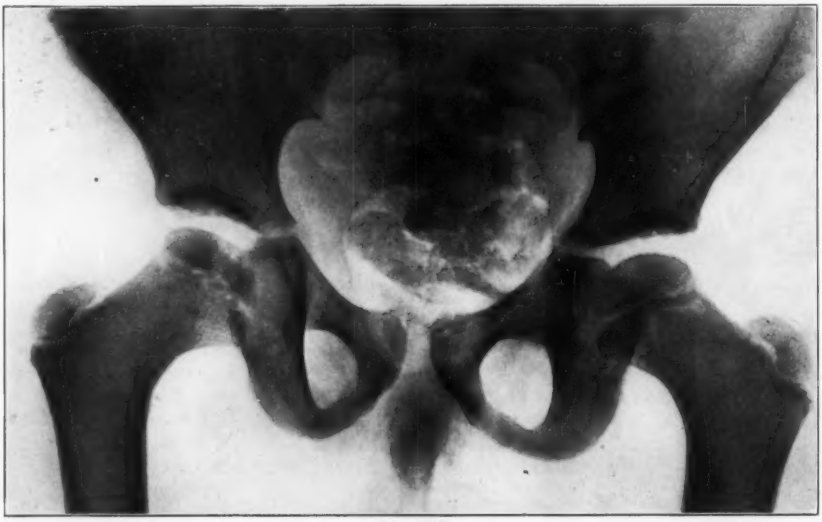

FIG. 7-B

FIG. 7-C

FIG. 7-D

A case with mild involvement and a satisfactory final result.

Fig. 7-A: Patient was first seen on July 5, 1945, at the age of six.
Fig. 7-B: December 29, 1945. Fig. 7-C: June 18, 1946.
Fig. 7-D: October 1, 1949. Four years after the patient was first seen, when he was ten.

CASE 5. An eight-year-old boy had had limp and pain in the right knee for six months. Moderate limitation of motion of his right hip was found. Roentgenograms (Fig. 5-A) revealed a large area of decreased density in the right metaphysis, surrounded by a thin line of increased density. Considerable lateral expansion of the flattened right femoral head was present. The articular cartilage space was widened. A diagnosis of moderately advanced Legg-Perthes disease was made and the patient was confined to bed for two years. He was then in a wheel chair for four months, and thereafter on crutches with partial weight-bearing for an additional three months. Roentgenograms made eleven months after bed rest had been begun (Fig. 5-B) showed that the lesion of the femoral neck was partially healed, but deformity of the femoral head and neck persisted. Roentgenograms made twenty-seven months after his first films (Fig. 5-C) demonstrated that the femoral neck was wide and somewhat shortened; replacement of the head was incomplete. At the age of twenty-three, the patient led an active life, but had occasional hip pain. The only significant finding was moderate limitation of rotation. Roentgenograms at that time (Figs. 5-D and 5-E) revealed an ununited

fragment at the superior aspect of the wide, flat femoral head. The femoral neck was shortened. Sclerosis of the right acetabular cortex was present.

The roentgenograms made when the patient was first seen demonstrated a large lesion in the neck of the femur and lateral expansion of the femoral head. In spite of prolonged bed rest, he had a poor final result.

Asymmetrical growth retardation or epiphyseal arrest is another factor which contributes to the development of deformity. In patients who obtained the best results, the epiphyseal cartilage plate always appeared to be normal and did not undergo premature closure. Patients with less satisfactory results often had evidence of growth retardation, and its effect was sometimes noted long after the femoral head had been replaced. The more rapid growth of the greater trochanter, which is unaffected, contributes to the

FIG. 8-A

FIG. 8-B

FIG. 8-C

FIG. 8-D

A poor result in a patient who had severe involvement.

Fig. 8-A: Patient was first seen on April 13, 1936, at the age of eight.
Fig. 8-B: December 28, 1936.
Fig. 8-C: June 21, 1937. Fig. 8-D: April 8, 1939.

distortion of the upper end of the femur. An example of asymmetrical growth retardation which produced deformity of the femoral neck is seen in Case 6.

CASE 6. An eight-year-old boy was seen because of slight limp and left groin pain of six weeks' duration. He had moderate limitation of left hip motion, accompanied by slight muscle spasm. Roentgenograms (Fig. 6-A) demonstrated a dense, mildly flattened left femoral head and a widened articular cartilage space. A diagnosis of early Legg-Perthes disease was made and, because of the acute symptoms and findings, traction was applied for one week. Crutches without weight-bearing were then used for twenty-one months. Roentgenograms (Fig. 6-B), made four months after treatment was begun, demonstrated partial replacement of the femoral head. Roentgenograms seven months (Fig. 6-C) and eleven months (Fig. 6-D) after the patient was first seen demonstrated lateral protrusion of the capital femoral epiphysis. A roentgenogram four years afterward (Fig. 6-E) showed that the femoral head had been completely replaced. However, early trabeculation across the lateral portion of the epiphyseal plate had occurred. There was moderate deformity of the femoral head and neck. Roentgenograms made at the age of twenty-one (Figs. 6-F and 6-G) showed the left femoral head to be somewhat flattened and widened. The femoral neck was asymmetrically short (Fig. 6-G).

The patient leads an active life, but has dull pain in the groin and thigh with exertion. He has 1.5 centimeters of shortening, 4 centimeters of thigh atrophy, and moderate limitation of rotation.

This patient had evidence of damage to the epiphyseal cartilage plate early in the course of his disease. The resultant flat, wide femoral head and asymmetrically short neck are probably the result of unequal growth retardation.

A comparative analysis of the best results and the worst results is seen in Table IV. Of the patients who had satisfactory results, sixteen obtained a nearly perfect hip. The majority of these children were under six at the onset of their disease. All but four of this group were treated early. In no case was there a severe disturbance of the neck. On the other hand, among the twenty-five patients comprising the worst group, the usual age at onset was three years later than in the preceding group. Furthermore, the majority of them either had no treatment or treatment was started late. One-fourth of them had extensive changes in the femoral neck.

FIG. 8-E
April 13, 1943

FIG. 8-F

FIG. 8-G

January 19, 1950. Fourteen years after treatment was begun, when the patient was twenty-two.

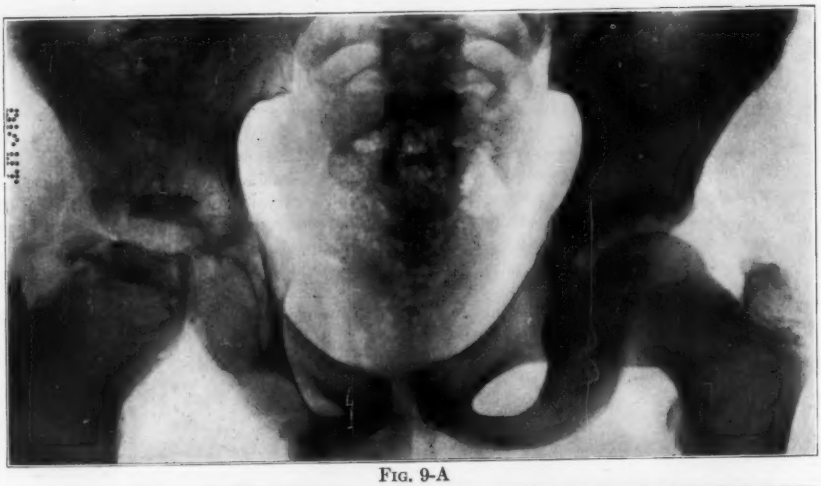

FIG. 9-A

FIG. 9-B

FIG. 9-C

FIG. 9-D

Severe involvement in another patient whose final result was poor.

Fig. 9-A: August 28, 1937. Patient was first seen at the age of ten.
Fig. 9-B: November 4, 1938.
Fig. 9-C: August 8, 1940.
Figs. 9-D and 9-E: March 24, 1949, when the patient was twenty-two.

FIG. 9-E

In several cases in the best group the entire course appeared to be quite benign, with little collapse of the head, no widening of the head and neck, and unusually rapid reconstruction. An example is Case 7.

CASE 7. A six-year-old boy was seen because of limp and pain in the right hip and knee of a few weeks' duration. Slight limitation of rotation was found. Roentgenograms (Fig. 7-A) revealed increased density and mild flattening of the right femoral head. A region of decreased density was present in the foveal area, where resorption had already occurred. A diagnosis of Legg-Perthes disease was made and non-weight-bearing on crutches was advised. The patient's family refused any treatment but did allow periodic examinations. Roentgenograms five months (Fig. 7-B) and eleven months (Fig. 7-C) after he was first seen showed that the femoral head was well replaced. Little fragmentation or collapse of the femoral head had occurred. The roentgenogram made at the age of ten (Fig. 7-D) revealed a normal appearing right hip joint. The patient was asymptomatic. Examination of the right hip was normal.

In contrast, the course of a severe disturbance is illustrated in Case 8.

CASE 8. An eight-year-old girl was first seen because of a slight limp and mild pain in her right thigh of four months' duration. The roentgenogram (Fig. 8-A) revealed a slightly flattened, dense, right femoral head. A diffuse area of rarefaction with a surrounding thin line of increased density was present in the metaphysis. The apparent subluxation of the right hip was due to the adducted and flexed position of the right thigh. A diagnosis of Legg-Perthes disease was made. The patient was then confined to bed for two years, during the first six months of which traction was applied. The roentgenogram made eight months after treatment was begun (Fig. 8-B) demonstrated thickening of the neck by new bone along its superior margin. New bone was also seen lateral to the femoral head. Roentgenograms after fourteen months of bed rest (Fig. 8-C) showed moderate resorption of the necrotic epiphysis. The bone lateral to the femoral head was more prominent. The roentgenogram seven years afterward (Fig. 8-E), when the patient was fifteen, revealed marked deformity of the femoral head and neck. Roentgenograms made fourteen years after the beginning of treatment (Figs. 8-F and 8-G), when the patient was twenty-two, disclosed a wide, irregularly

overgrown right femoral head, a large portion of which was outside the acetabulum. The acetabular cortex was somewhat wide, sloping, and irregular. The right femoral neck was asymmetrically short and a loose body was present at its posterior aspect (Fig. 8-G).

This patient, who is very obese, stands all day at her work and by nightfall a mild hip flexion contracture is present. She has pain in her right thigh as a result of exertion or weather changes. Considerable limitation of rotation and abduction and moderate instability of her left hip are present.

The preceding patient was fairly old at the onset of her disease. Treatment was not begun early. During the course of her disease, a distinct neck lesion developed, lateral expansion of the femoral head, and new-bone formation along the superior border of the femoral neck. As would be expected from these findings, this patient has a poor result.

Case 9 concerns another patient whose outcome was poor. This patient was even older at the onset, and his disease was moderately advanced before treatment was begun. A central depression of the necrotic femoral head developed. Most cases with similar depressions have poor results. New bone developed along the superior margin of the femoral neck and contributed to the deformity. The early fusion of the capital femoral epiphysis is evidence of damage to the epiphyseal cartilage plate.

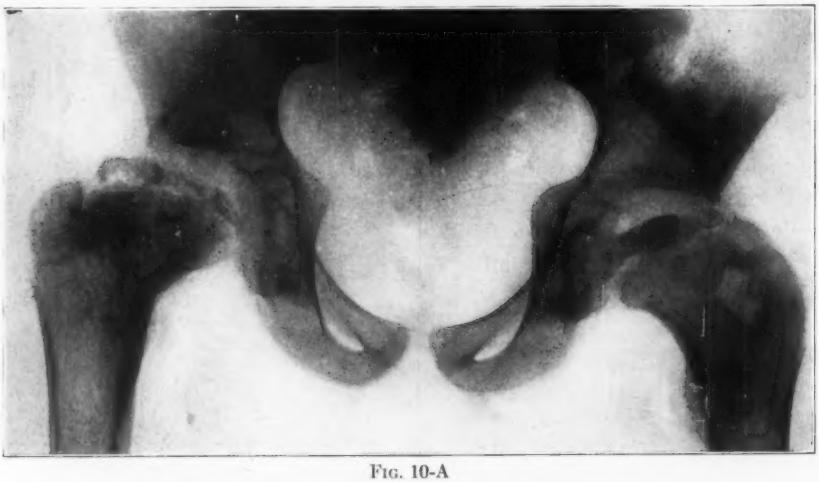

FIG. 10-A

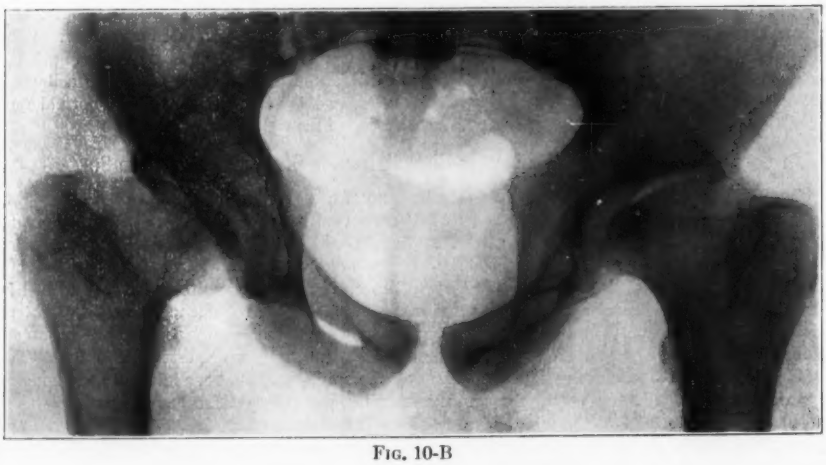

FIG. 10-B

CASE 9. A ten-year-old boy was seen because of limp and pain in his right groin for nine months. Muscle spasm, pain, and marked restriction of motion were present. The roentgenogram (Fig. 9-A) revealed areas of increased and decreased density in the right femoral head. A depression of the central portion of the dense femoral head was seen. This had the appearance of a fracture. The articular cartilage space of the right hip was widened. A diagnosis of moderately advanced Legg-Perthes disease was made. The patient was confined to bed for eight months and was then in a wheel chair or on crutches with weight-bearing for four months. The roentgenogram fifteen months after treatment was begun (Fig. 9-B) revealed a shortened, thickened femoral neck with new-bone formation on its superior aspect. The foveal area of the femoral head had not yet been replaced. Roentgenograms three years afterward, at the age of thirteen (Fig. 9-C), showed that the right femoral capital epiphysis was practically closed while the left remained open. The roentgenograms made at the age of twenty-two (Figs. 9-D and 9-E) revealed the femoral head to be thickened and widened. An exostosis was present on the anterior surface of the neck, near the site of the old epiphyseal plate. The right acetabular cortex was slightly denser than the left.

This patient, who does heavy work as a carpenter, occasionally has sharp pain in the anterior aspect of his right hip and a slight limp. He has 5 centimeters of thigh atrophy and moderate limitation of motion of the right hip.

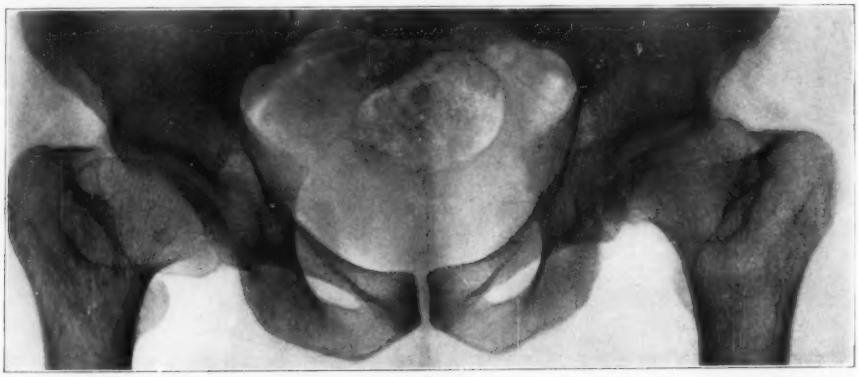

FIG. 10-C

FIG. 10-D

FIG. 10-E

A poor result in an untreated patient who was followed into middle life.

Fig. 10-A: The patient was first seen in 1924, at the age of eleven.
Fig. 10-B: February 1929.
Figs. 10-C, 10-D, and 10-E: January 7, 1950. Twenty-six years after the patient was first seen, when he was thirty-seven.

It is probable that some of these patients will in later life have more disability. Similar patients who are now in middle life do have more symptoms, as the following two cases show.

CASE 10. An eleven-year-old boy had had limp and pain in his left hip for one year. The roentgenogram (Fig. 10-A) revealed changes of bilateral far advanced Legg-Perthes disease. The right femoral head had been almost completely replaced, but was markedly widened and flattened. The right femoral neck was thickened and shortened. The left femoral head was also flattened and widened, and a large area of increased density persisted within it. The left femoral neck was shortened and contained a large area of decreased density. Biopsy of this lesion was performed. The patient refused treatment. A roentgenogram four years

FIG. 11-A

FIG. 11-B

Another poor result in an untreated patient who was followed into middle life.

Fig. 11-A: March 31, 1916. The patient was first seen in 1914, at the age of four.
Fig. 11-B: July 6, 1917.
Figs. 11-C and 11-D: December 17, 1949. Thirty-five years after the patient was first seen, when she was thirty-nine.

TABLE II

CORRELATION OF LATE RESULTS WITH TREATMENT IN SEVENTY-THREE CASES

Type of Treatment	Results			Total
	Satisfactory	Fair	Poor	
None	3	1	9	13
Early				
Ambulatory	19	4	2	25
Non-ambulatory	14	4	3	21
Late				
Ambulatory	4	1	2	7
Non-ambulatory	1	1	5	7
Total	41	11	21	73

later (Fig. 10-B), at the age of fifteen, demonstrated the marked deformity of both hips. Roentgenograms made when the patient was thirty-seven (Figs. 10-C, 10-D, and 10-E) showed the irregular femoral and acetabular cortices, most marked in the left hip. Both femoral necks were shortened and deformed. The greater and lesser trochanters were higher than normal.

This patient does heavy work, and he loses some time from his job because of severe pain in the left hip, associated with changes in the weather. He has marked limitation of rotation, flexion, and abduction in both hips and a constant limp.

CASE 11. A four-year-old girl was first seen because of right hip pain. Roentgenograms made two years later (Fig. 11-A) revealed flattening, widening, and irregularity of the right femoral head with a widened articular cartilage space. The right femoral neck was thickened. A diagnosis of Legg-Perthes disease was made, but the patient was not treated. Sixteen months later (Fig. 11-B) the femoral head had been partially replaced. The head and neck had widened further. Roentgenograms made thirty-five years after she was first seen (Figs. 11-C and 11-D), when the patient was thirty-nine, demonstrated the markedly enlarged, flattened, and overgrown right femoral head. The femoral neck was shortened.

FIG. 11-C

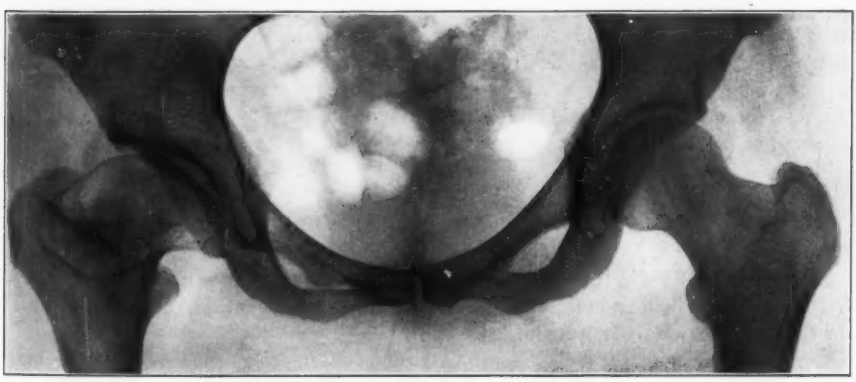

FIG. 11-D

The patient now has a constant limp, moderate instability, and considerable pain in the right hip. If she were not of such a slight build and did not lead a sedentary life, she would probably have more disability.

TABLE III

CORRELATION OF LATE RESULTS WITH AGE AT ONSET IN SIXTY-TWO TREATED CASES

Age (*Years*)	Results			Total
	Satisfactory	Fair	Poor	
Under 5	10	0	1	11
5 to 8	27	6	7	40
Over 8	2	5	4	11
Total	39	11	12	62

TABLE IV

COMPARISON OF THE BEST RESULTS WITH THE WORST RESULTS

Type of Result	No. of Cases	Average Age at Onset (*Years*)	Treated Early (*Per cent.*)	Severe Neck Lesions
Poor	25	8.1	24	6
Nearly perfect	16	5.0	75	0

DISCUSSION

The prognosis in a case of Legg-Perthes disease can in a general way be estimated early in its course. Younger patients have been shown to have a higher incidence of satisfactory results. Evidence of severe femoral-neck disturbance with damage to the epiphyseal plate is followed by deformity and a poor result. Often only a portion of the epiphyseal cartilage is damaged and the result is asymmetrical growth retardation, which may not become evident for some time. This agrees with the studies of Gall and Bennett and others, who had the opportunity of studying necropsy material [21]. They found evidence of marked irregular damage to the epiphyseal plate with complete breakdown and destruction in many areas.

Inasmuch as no significant difference is found in the results from non-ambulatory and ambulatory methods of treatment, the latter is considered the treatment of choice when the disease is unilateral. Occasionally, when the initial symptoms and findings are marked, bed rest or traction for a week or two is beneficial. However, complications from prolonged bed rest and immobilization do occur. The use of crutches avoids these complications and permits a fairly normal existence. For the younger child, a halter attached to the shoe maintains knee flexion; only rarely is it necessary to provide more restriction or to resort to hospitalization for an uncooperative patient.

CONCLUSIONS

1. In the natural course of Legg-Perthes disease there is a wide variation in the severity of the process.

2. Marked changes in the femoral neck and lateral expansion or central depression of the femoral head indicate a bad prognosis. In general, the younger the patient at the onset of the disease, the better the prognosis.

3. Premature symmetrical or asymmetrical growth arrest of the capital femoral epiphyseal cartilage plate may occur and contribute to the deformity.

4. In the presence of gross changes on the roentgenograms, there may be few or no symptoms in early adult life. However, as the patient becomes older, progressive disability does occur.

5. Therefore, patients should be treated and treated early. Inasmuch as there seems to be no significant difference in the results obtained by ambulatory or non-ambulatory treatment, non-weight-bearing on crutches is sufficient in a cooperative patient with unilateral involvement.

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THE CURE OF EXPERIMENTAL SCOLIOSIS BY DIRECTED GROWTH CONTROL *†

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The development of techniques for the production of spinal curvature in animals has paved the way for experiments on the correction of such deformities. Of the various procedures used to produce scoliosis [1, 4, 6, 9, 12, 15], many have been criticized on the ground that they yield distortions which are not analogous to those of idiopathic scoliosis,—that they simulate the angulations seen in certain fractures or in congenital anomalies. One method, however, yields curvatures which are singularly like the human deformity [14]. This present paper is a report on the cure of curvatures so produced.

The experimental production of these curvatures is based on the recognition of four facts: (1) that vertebral bodies grow in height by the activity of their epiphyseal cartilages [5, 11], (2) that epiphyseal growth can be retarded by compression [2, 3, 7, 10, 13, 16], (3) that the power of growth persists after the compression is removed [2, 8], and (4) that unequal elongation of the two sides of the spine will result in spinal curvature. These principles have been applied by the unilateral clamping of several lumbar vertebrae in growing dogs [14]. The clamp is so inserted that the epiphyses are not injured (Fig. 1-A). The compression force is developed by the normal growth of the spine against the restraint of the clamp. In puppies so prepared, lateral curvature develops after six to eight weeks, and the deformities are remarkably like those seen in children. If fixed lateral curvature of the spine, rotation of the vertebrae, and absence of roentgenographic evidence of disease constitute the criteria for the diagnosis of scoliosis, these dogs may be said to have scoliosis. They also show such additional characteristics as secondary curves and torsional changes in the thorax. Roentgenographically, they present wedging of the vertebrae and unilateral narrowing of the intervertebral spaces (Figs. 1-B and 1-C). If the clamp is removed after the curve has been established, the deformity persists or even increases (Fig. 1-C). The secondary curves, which represent adaptation changes and which are not produced by surgical tampering with the vertebrae involved in their curvature, deserve attention. They offer the opportunity to study compensatory mechanisms. Their behavior in response to the straightening of the experimental primary curves may prove of value in understanding human scoliosis.

EXPERIMENTS

In the search for a laboratory method to cure scoliosis, the proposition presented itself that, if selective growth control can make a straight spine grow curved, it might also make a curved spine grow straight. To test this hypothesis the following experiments were undertaken:

Six dogs with fixed rotatory lateral curvature of the spine were subjected to selective growth control. The animals were still in the growing age. Their deformities had been produced by the stapling technique here described. The stability of these deformities was tested as follows: Varying periods of time were allowed to elapse between the curve-producing and the curve-correcting phases of the experiment. However, time was limited, since there was danger of losing the growth potential. Nevertheless, in at least one animal, six weeks was allowed to elapse (Figs. 2-A and 2-B). No evidence of spontaneous cure was observed in these animals. To determine the course of the curvatures over longer

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FIG. 1-A

FIG. 1-B

FIG. 1-C

FIG. 1-D

X-rays of a dog's spine to show effects of unilateral stapling and persistence of the deformity after removal of the staple.

Fig. 1-A: On the day of stapling.

Fig. 1-B: Seven weeks later, one day prior to removal of staple. Lateral deviation and rotation are marked. Unilateral wedging of intervertebral cartilage is noted.

Fig. 1-C: Nine months later, after normal freedom without corrective treatment. Film shows right thoracic, left lumbar curvatures with rotation, thoracic distortion, and characteristic wedging.

Fig. 1-D: Photograph of skeleton of dog in Figures 1-A, 1-B, and 1-C. (Evaluation of lateral displacement is aided by squaring up the pelvis.)

FIG. 2-A FIG. 2-B FIG. 2-C FIG. 2-D

X-ray of dog's spine to show effects of unilateral growth control.

Fig. 2-A: One month after stapling together the fourth, fifth, and sixth lumbar vertebrae.

Fig. 2-B: Six months after stapling and six weeks after removal of staple, showing a left lumbar curvature with a compensatory right thoracolumbar curve.

Fig. 2-C: Immediately after applying corrective stapling on the convex side. The staple is applied to the third, fourth, and fifth lumbar vertebrae, leaving the wedged intervertebral cartilage below the fifth lumbar vertebra to serve as a control.

Fig. 2-D: Five months later, on removal of corrective staple. The wedging below the fifth lumbar vertebra persists; the segment that was stapled is straightened. The secondary curves are less pronounced. Below the sixth lumbar vertebra, a mild compensatory curve is noted.

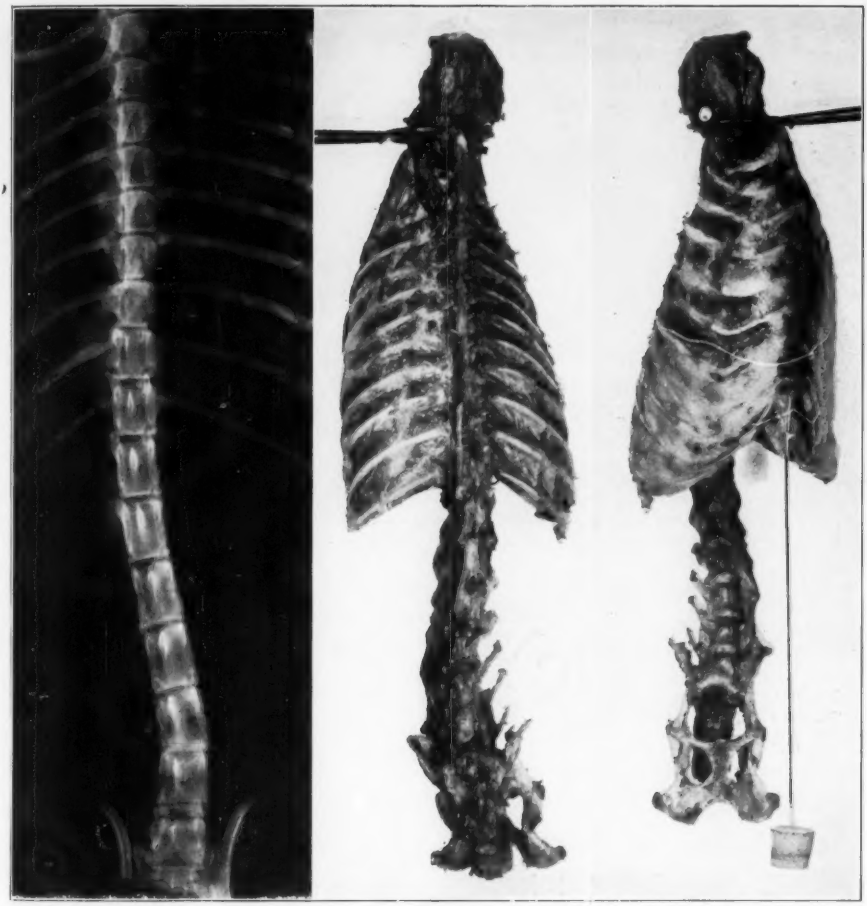

FIG. 2-E FIG. 2-F FIG. 2-G

Same animal as seen in Figures 2-A, 2-B, 2-C, and 2-D, after autopsy.
Fig. 2-E: X-ray of skeleton.
Fig. 2-F: Photograph of skeleton. Posterior view.
Fig. 2-G: Photograph of skeleton. Anterior view.

stretches of time, several dogs with established curves were freed of their staples and were sent out to live in kennels. They were observed for periods up to eight months, but in none of these animals was the curvature affected. In one puppy, the corrective clamp was so placed that it did not encompass all of the epiphyses originally stapled. Instead, the lower two wedged epiphyses in the induced curvature were allowed to remain without growth restraint, while the epiphyses of the three higher vertebrae were clamped. In this animal, the unrestrained lower deformity persisted even though the upper curvatures were favorably affected by the staple (Figs. 2-C and 2-D). One may therefore reasonably assume that the corrective changes noted in the deformities of the experimental animals would not have appeared without active intervention.

PROCEDURE

The treatment used consisted in clamping together the vertebrae in the primary curve on the convex side. With the animal in the prone position, a paraspinal incision

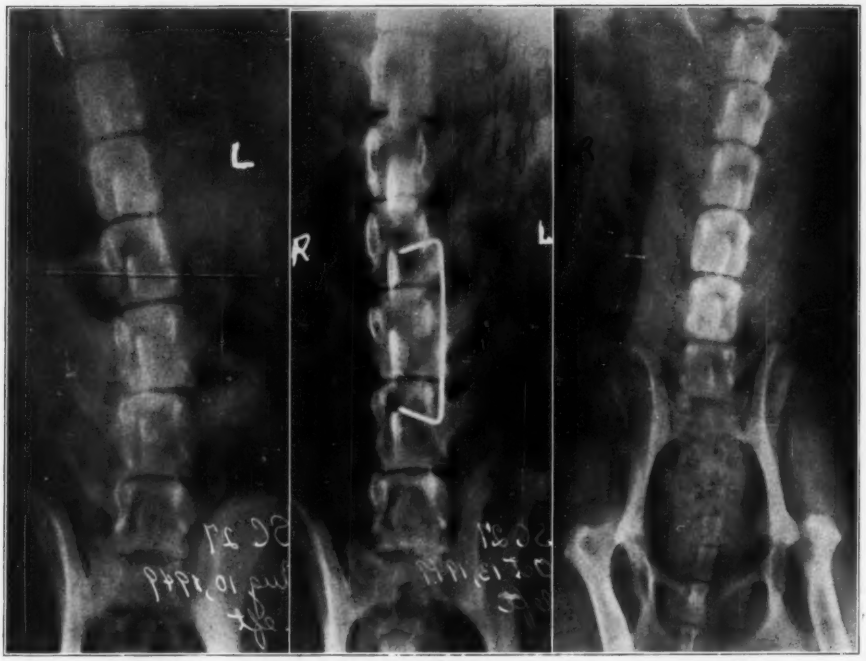

FIG. 3-A FIG. 3-B FIG. 3-C

Fig. 3-A: Three months after stapling fourth, fifth, and sixth lumbar vertebrae and four weeks after removal of staple a left lumbar curvature persists.

Fig. 3-B: Six weeks after application of corrective clamp, spine is practically straight.

Fig. 3-C: Six months after corrective clamp was applied and three months after its removal, spine shows overcorrection of curves.

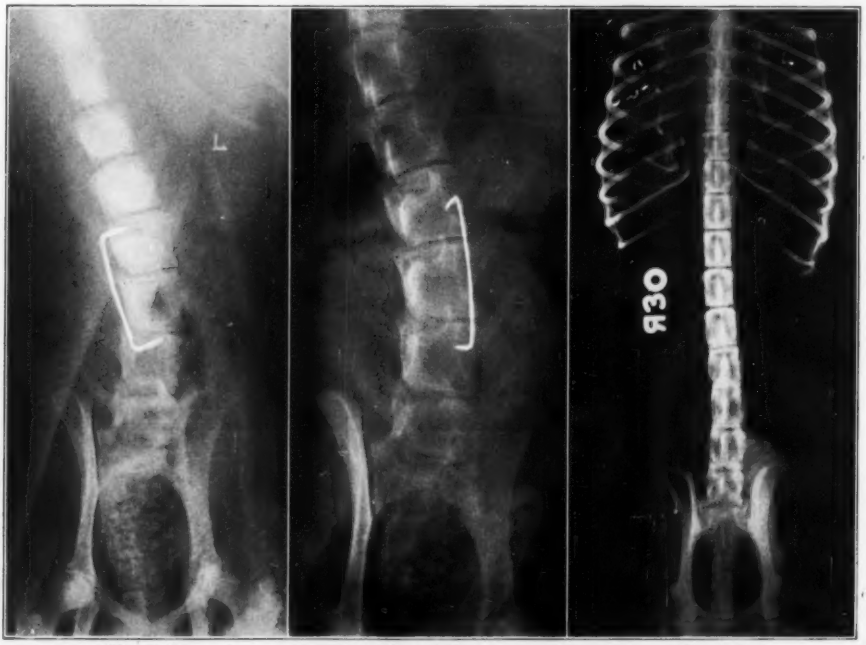

FIG. 4-A FIG. 4-B FIG. 4-C

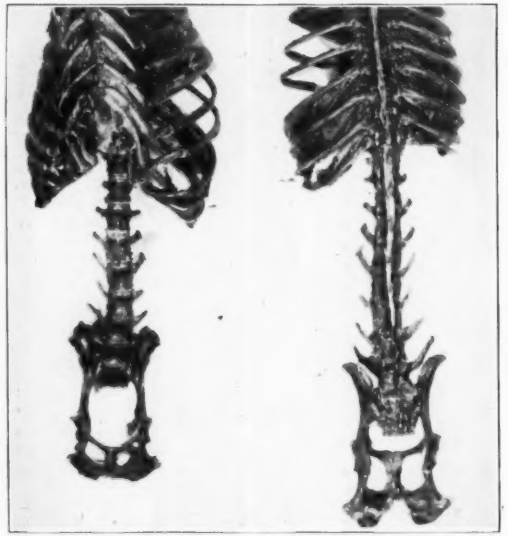

FIG. 4-D FIG. 4-E

Fig. 4-A: Four months after stapling, curvature is present.

Fig. 4-B: One week after corrective stapling with spine forced straighter.

Fig. 4-C: After autopsy, five and one-half months after application of corrective treatment.

Figs. 4-D and 4-E: Photograph of skeleton of animal in Figures 4-A, 4-B, and 4-C.

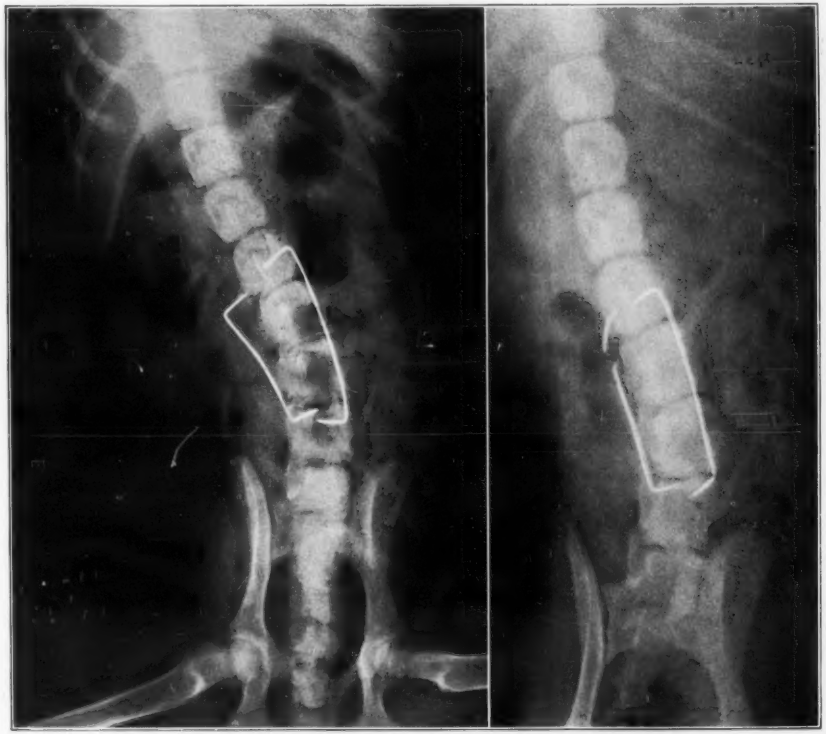

FIG. 5-A FIG. 5-B

X-rays of a dog's spine.

Fig. 5-A: Original staple applied on right side three months before seems to be pulled out. Corrective staple on left has just been applied.

Fig. 5-B: Five months later, both primary and compensatory curves are less pronounced. (Original staple has broken.)

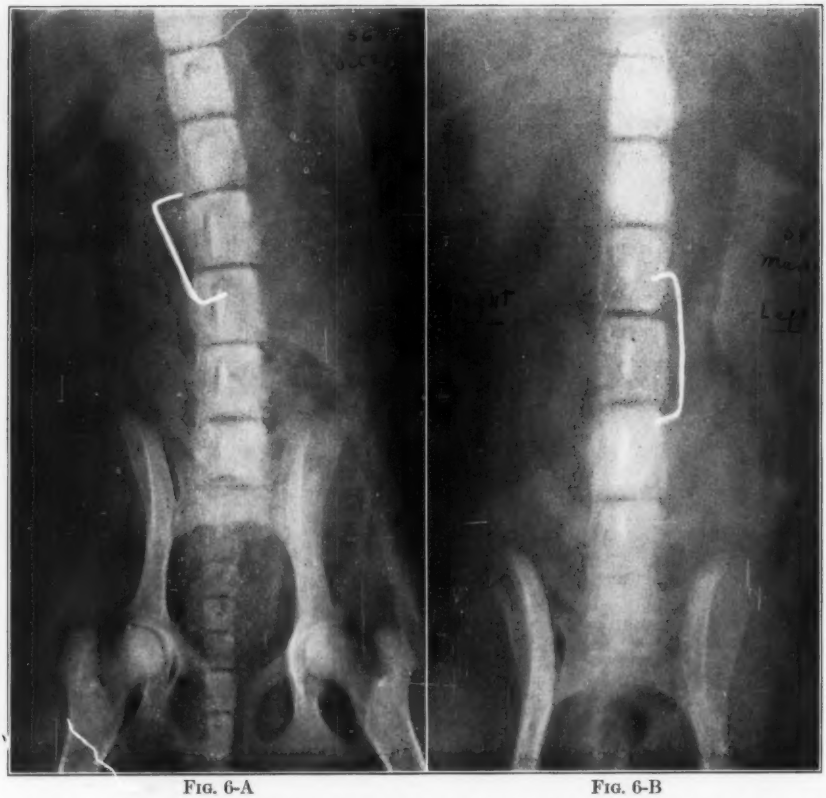

FIG. 6-A FIG. 6-B

Fig. 6-A: Ten weeks after stapling, clamp is found to have pulled out, but moderate deformity is present.

Fig. 6-B: Four months after corrective stapling spine is straight.

was made in the lumbar area on the convex side of the spinal curve and was deepened to expose the sacrospinalis muscle. This was split bluntly over a line connecting the tips of the desired transverse processes. Periosteal elevation medially brought to view the junctions of these processes with the neural arches. Here a perforator was driven anteriorly and medially in the predetermined vertebrae; and the prongs of a staple were introduced into the prepared holes, so that the bodies of the vertebrae were entered without damage to the epiphyses. Closure of the soft tissues was then made in layers, and after a short postoperative recovery period, the animals showed no ill effects or disabilities attributable to the operation.

RESULTS

In the six deformed dogs, which had treatment by selective growth control, all showed some correction. In two, the spines became practically straight (Figs. 4-A to 4-E; 6-A and 6-B); in two, appreciable improvement took place (Figs. 5-A and 5-B; 7-A and 7-B); in one, there was overcorrection (Figs. 3-A, 3-B, and 3-C); and in one, treated by an experimental variant, the segment under growth control became straight (Figs. 2-C and 2-D).

The behavior of the secondary curves is most interesting. In the two "cured" animals, without special local treatment, the compensatory curves cleared up concurrently

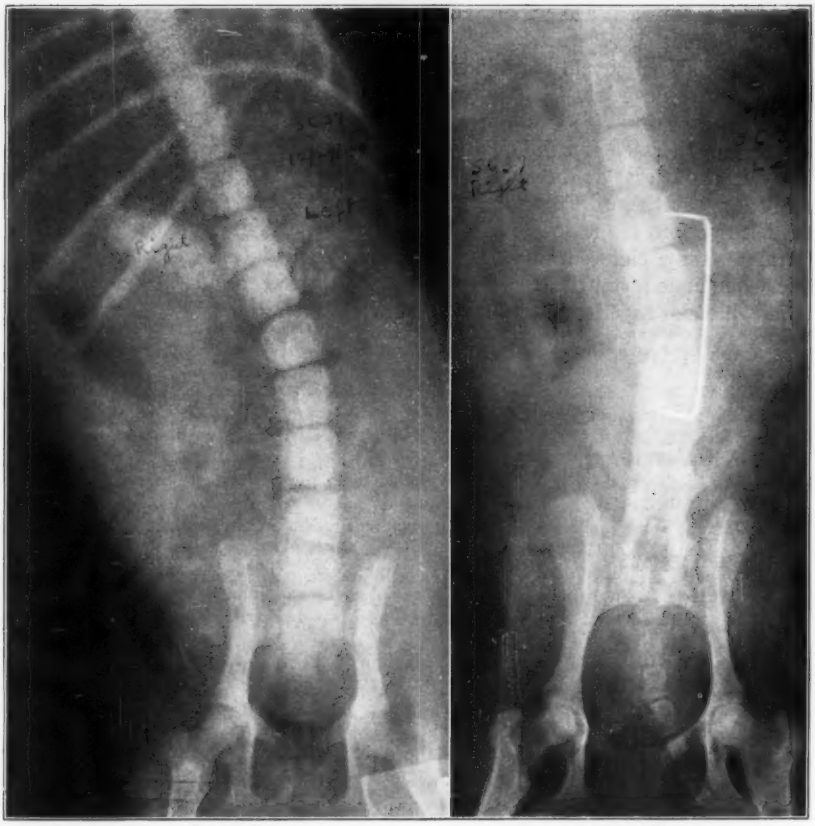

FIG. 7-A FIG. 7-B

X-rays of dog's spine.

Fig. 7-A: Curvature following seven weeks' stapling from the second lumbar vertebra to the fifth lumbar vertebra on the right side.

Fig. 7-B: Two months after corrective stapling clamp is pulled out, but spine is straighter.

with the realignment of the lumbar vertebrae. In the animal whose lumbar curve was overtreated, so that the left lumbar deviation became a right lumbar curve, the secondary thoracolumbar deviation shifted from the right side to the left.

It is evident that the progress of the corrective treatment must be closely watched. Our case of overcorrection was not due to an oversight, but was anticipated. On the other hand, the incomplete correction in two cases resulted from premature termination of the experiments in anticipation of continued epiphyseal growth in the right direction. It is probably wise to remove the staples when the spine is practically straight, if one desires full correction.

DISCUSSION

On the basis of the production of rotatory lateral curvatures in twenty-three normal growing dogs by unilateral lumbar growth restraint, the development of a similar deformity in one animal with thoracic stapling and the correction of deformed spines under selective growth control in the six dogs just reported, it is obvious that a tool is available for the making and straightening of spine deformities. This assertion must at present be limited to puppies, since the operations were performed in dogs only. Yet, if the premises

on which this experiment was undertaken hold true in other vertebrates, one may expect similar results in other groups.

Thus, the human should react in like manner to directed growth control, since his vertebral bodies gain height by epiphyseal structures [5, 11], and since the epiphyses in his extremities show growth retardation under compression. There is much to be learned about the growth rates of children's vertebrae, and some technical changes would be necessary in the operation, because of the differences in the shapes of the human and canine bones, but these difficulties are not insurmountable. Indeed, the operation has already been performed on three children and has proved to be simple and non-shocking. Furthermore, these children have shown remarkably little discomfort or limitation of activity while undergoing this treatment. A report of end results, however, must await more time, broader experience, and further study.

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DISCUSSION

Dr. John R. Moore, Philadelphia, Pennsylvania: Dr. Nachlas has presented a most interesting paper. Both he and Dr. Borden are to be congratulated upon this very fine piece of original work. A rotatory lateral curvature in a dog has been described. Dr. Nachlas believes that this is identical with the lateral rotatory curvature seen in the human. Apparently he believes that all lateral rotatory curvatures of the human are due to epiphyseal disturbance. There is undoubtedly epiphyseal-plate disturbance occurring in any type of scoliosis, which has its inception in the growth period. The etiology may be contracted pleurae, fusion of the ribs, muscle imbalance secondary to poliomyelitis and other lower motor neuron lesions, trauma, *et cetera*. Does he believe that arresting the epiphyseal growth in the case of a scoliosis produced by severe pleural contracture or a severe burn scar, will bring about a correction of the scoliosis? The placement of the staples for the correction of a scoliosis and for that matter, for the production of a scoliosis as observed in his experimental work, is relatively simple. However, where would the staples be placed to correct a scoliosis in which there is 60 degrees of rotation and a correspondingly severe lateral curvature? Could the

same axis be employed which is used in his experimental work? This method, if proved to be of practical value, will have to be restricted to very early curvatures, and it is quite likely that it would be necessary to correct these curves to neutral before the application of the staples. From the experience I have had with the stapling of the transverse processes in hope of producing a growth arrest on the same side, I found it not only necessary to produce the maximum correction but also necessary to hold the spine in this corrected position with every power at my disposal in order to prevent the spine from springing back and becoming unstapled.

DR. ARTHUR STEINDLER, IOWA CITY, IOWA: What appears important to me in this most interesting paper of Dr. Nachlas's is not so much the technique which he applies to arrest vertebral growth but is his efforts to produce and control scoliosis which are based upon the growth element rather than upon muscle imbalance.

It was on the grounds of muscle deficiency that the experimental production of scoliosis was attempted by earlier observers. Wullstein tried to produce it by strapping animals in scoliotic positions, and Schwartzmann and Miles tried to obtain it by unilateral stripping or excising muscles. We must recognize today that none of these experiments were able to produce a true scoliosis by the muscle imbalance alone; when it did occur, it was a secondary growth phenomenon. It is becoming more and more evident that true scoliosis cannot be produced experimentally unless the experiment also destroys what we call the intrinsic equilibrium of the spine, by damaging the epiphyseal plates and the intervertebral discs.

I call that element in the scoliosis which is due to muscle inbalance, either primary or secondary, the inclinatory or contractural element; the other is the so-called collapse element. This embraces the loss of the intrinsic equilibrium of the spine due to degenerative changes of the intervertebral discs, of the vertebral ligaments, or to disturbance of the normal epiphyseal growth. The important point is, that it is only the contractural element which yields to the ordinary method of correction by corrective casts — and that, only within very narrow limits; so that the effect of correction is the development of counter-curves rather than a straightening of the original curve, itself. So far as the collapse element is concerned, it is entirely based on degenerative changes of non-contractile structures, and the degeneration of the intervertebral disc. We may just as well realize that nothing can influence it, unless it is such a method as that indicated in Dr. Nachlas's paper, which makes a direct attack upon the growth mechanism of the vertebrae.

This distinction between the conservatively still-manageable contractural element, and the growth element, which can be affected only by the methods Dr. Nachlas described, or similar ones, should, I believe, control our entire philosophy regarding the treatment of scoliosis. It is true that stapling may not be the only way to influence growth inequality. We all remember the work of Haas on vertebral epiphyses. Engel and Richer have described experimental production of scoliosis by asymmetrical radium irradiation. Many others have done work on similar lines.

I think I can safely conclude my remarks with the statement that, because of the principle involved, Dr. Nachlas's paper is of importance, not only regarding the pathogenesis of this deformity, but also because it opens up certain possibilities for correction.

DR. ARTHUR B. LEMESURIER, TORONTO, CANADA: I have had no experience in controlling the growth of the spine by stapling, but I have had some experience in treating scoliosis by attempting to control the growth on one side only by damaging the epiphyseal cartilaginous plates of several vertebral bodies. This idea was suggested by two papers, one by S. L. Haas, published in 1939, and the other by J. Dewey Bisgard and M. M. Musselman, published in 1940. In both of these, a somewhat similar method was described of producing scoliosis in animals by damaging the epiphyseal cartilaginous plates on one side only, and in both it was suggested that the method be used to treat scoliosis in human beings. Between the years 1940 and 1942, I operated on four children in this way. They all seemed to be ideal cases for this method in that they all had congenital scoliosis with a single hemivertebra in the lumbar region. There was no doubt as to which was the primary curve, and in this region the sides of the vertebral bodies were accessible through the incision described by Royle for lumbar sympathectomy. Further, the patients were all between the ages of three and five years, with enough growth still to take place to show the effect of the operation.

The same operation was performed in all four cases. The convex side of the vertebral column was exposed in the lumbar region, and the six epiphyseal cartilages of three vertebrae, including the wedge-shaped one, were destroyed. The destruction was done by a knife, an osteotome, a curette, and a cautery; and extended as far as the mid-line of the vertebral body. The destruction of the growth cartilage and the bone adjacent to it seemed to be complete. Since then, all have been followed for a period now of eight or ten years. The vertical height of all the vertebral bodies operated on has increased considerably in that time, and this increase has taken place to the same extent on the side operated upon as on the undamaged side. Apparently, the operation, as done, had no appreciable effect on the growth. There has been no improvement in any of the curves. As a matter of fact in all the cases the deformity has increased, mostly in the two or three intervertebral spaces above the hemivertebra, and two of these children have since had their spines fused.

It is possible that the destruction to the epiphyseal plates in these cases might have been better done, but the results in all four were so disappointing that we have not performed the operation again since 1942.

Dr. Nachlas's method of controlling the growth of one side of the spine by stapling is probably more efficient than the method of destroying the epiphyseal plates has proved to be. It is not likely to be the answer for all cases of scoliosis, particularly for the idiopathic cases, but it certainly warrants further investigation. No method at present available for the treatment of scoliosis can be considered adequate since we have to be satisfied with a result that is far from perfect. Any new method that holds out hope of improvement should at least be considered and, if it is within the limits of safety, should be encouraged.

Dr. John R. Cobb, New York, N. Y.: This is a very interesting experimental study. The one thing that I would object to is its application to humans at this time. Dr. Nachlas has shown by these experiments what we all would expect from the work done by Dr. Blount in stapling other growing areas.

I would like to discuss the application of this stapling procedure to human patients. Many of us will admit that we have had trouble with procedures for spine fusions. Our results have not always been good; we sometimes have complications when we do a spine fusion, which is a relatively simple operative procedure compared with the stapling of the vertebral bodies in the thoracic area in a young child. Perhaps the complications of this operative procedure, if it is accepted too quickly in its application to humans, might be serious.

We should first consider a fundamental principle. Many have assumed that growth is the cause of the increase in the curvature because the increase occurs during the growth period. We must be careful to distinguish, therefore, between a concomitant occurrence and a causative factor. We all know that in most cases of idiopathic scoliosis the curve does not increase to a severe degree with or even without treatment. I have followed many patients from the age of eight, ten, or twelve to the age of fifteen, sixteen, or older, who have had no treatment and their curvatures have not increased. Some of these patients have increased their height by as much as six, ten, or even twelve inches. Before any claim can be made for the efficacy of any treatment for any condition, we must know what happens in a large series of cases without any treatment. Unless we can be shown that this method, or any method of treatment, produces better results than those obtained without any treatment, it will be difficult to prove that arrested growth control, or any other form of therapy, is the factor in preventing the increase in these cases.

Since idiopathic scoliosis stops progressing spontaneously at an unpredictable time,with or without treatment, it is difficult to prove that any treatment in any specific case was the factor that prevented its increase. I would like to emphasize that dangers are involved in applying this operation to humans, and perhaps, since we have been quoting some of the literature, we might quote Alexander Pope who said two hundred years ago:

"Be not the first by whom the new are tried,
Nor yet the last to lay the old aside."

Dr. Paul B. Steele, Pittsburgh, Pennsylvania: In 1922, we tried to produce the mechanics of scoliosis in dogs by taking out the articular facets and inserting a wedge which lifted up one side of the vertebra. We took out two or three facets at the first operation and two or three at another operation. However, we found that, for our purposes, the facets of a dog were a little too small; therefore, we went into the laboratory and produced scoliosis on cadavera.

In 1924, we performed our first operation on the articular facets in a human being with scoliosis. Up to the present time, we have done some thirty operations in which we have taken out either two or three facets in the lumbar region. None of these operations was done on a patient under fourteen years of age. In an attempt to correct the torsion and rotation of the vertebrae and, at the same time, to lift the lower side up, we took out the articular facets on both sides and lifted the narrow side by inserting wedge-shaped chips of beef bone, which maintained the correction while the bone, turned down from the lamina, was fusing. After the operation, all patients were immobilized in the overcorrected position with a spica and body cast for three months. Three months is usually the time allowed to give assurance that fusion has taken place. Since we get permanent correction at the time of operation, we allow the patients up and give them no further treatment other than a few exercises at the end of the three-month period.

About three months ago, we saw a fourteen-year-old girl on whom we had recently performed this operation. On examination, we found her spine to be perfectly straight, much to our surprise, for usually patients who have undergone this operation have a little resulting deformity, which may decrease as they grow.

In older patients who have very marked scoliosis with a pronounced deformity, in which the ribs are in the pelvis, it is also possible to get a positive correction at the time of operation. One such patient, a woman who had had the ribs lifted out of the pelvis, now requires no brace.

CONGENITAL ANOMALIES OF THE HAND *

BY ARTHUR JOSEPH BARSKY, M.D., NEW YORK, N. Y.

This paper is based upon our own series of sixty-two cases—most of them, although not all, seen during the last five years—and a survey of the medical literature for the last thirty years. Over one hundred articles were reviewed, as well as the standard works on paleopathology and six Egyptian papyri. For the most part the contemporary articles consisted of case reports or genealogies, and in some of them the literature pertaining to a particular anomaly was reviewed. Comparatively few general articles were found. Recently a monograph by Birch-Jensen of Denmark has been published, dealing principally with genetic and statistical aspects of the subject.

We were interested in finding the earliest possible references to congenital anomalies of the hand; but, although a careful search was made, no definite information was discovered in the works of the paleopathologists. We surmise that this is due to the almost impossible task of collecting and assembling the small bones of the hand. Our observations along these lines confirm those of Elliot Smith and F. Wood Jones, who reported an archaeological survey of Nubia in which thousands of skeletons were excavated, and who remarked on the difficulty of handling the small hand bones.

The thought occurred to us that the examination of mummies, dating back to about 3000 B.C., whose entire bodies had been preserved, might perhaps yield some information. Here again we were unsuccessful. Although several congenital anomalies were reported in mummies, there is no mention of hand conditions. This failure may be accounted for by the fact that, since most mummies are of the royalty, the custom of brother-sister marriages may have prevented the introduction of outside hereditary factors. A final possible source of information—Egyptian art—also failed to give information about the period. However, we feel that the ancients, no less than the moderns, were undoubtedly afflicted with anomalies of the hand, even though no actual evidence was found to support this view.†

The earliest written reference that we have discovered is in the Bible, which refers to the giant of Gath as having six fingers and toes. This period has been fixed at about 1000 B.C. One of the most remarkable pedigrees on record is that of hexadactylism in the Scipian family, transmitted for a recorded seven hundred years. Another interesting historical incident was reported by Drinkwater in 1917, who traced a history of symphalangism to the time of Henry VI. The first member of the family known to have had the condition was killed in 1453. In 1874 his tomb was opened and identified, and his finger bones showed the same ankylosis as that displayed by his later descendants.

We have been unable to find any comprehensive statistics of the occurrence of congenital hand anomalies which seem valid. The most detailed studies of congenital malformations, those of Murphy, unfortunately do not specify either the number or the percentage of hand involvements. Birch-Jensen's recent monograph, a carefully recorded study of all the living patients in Denmark, tabulates 625 patients with hand anomalies, or an average of one per 6,438 persons. However, this study does not include polydactylism or simple uninvolved syndactylism, our most frequent anomalies; therefore, it is not possible to apply the figures accurately.

The etiology of congenital anomalies of the hand has not been conclusively established, but at the present time the most acceptable theories are those of maldevelopment or of mutations which are subsequently inherited. A mutation is defined as a permanent

* Presented at the Fifth Annual Meeting of the American Society for Surgery of the Hand, New York, N. Y., February 10, 1950.

† The neolithic cave paintings in the Pyrenees are conceded to show mutilations, not congenital anomalies.

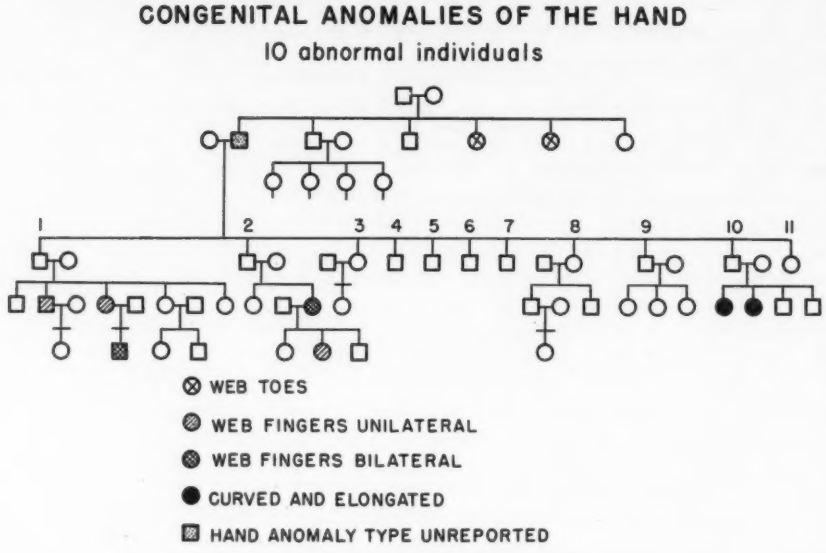

FIG. 1

Chart of the "X" family, showing the manifestations of hereditary congenital anomalies of the hands and feet. The pattern is not that of a Mendelian dominant, nor does the same anomaly appear throughout the generations. However, there is a definite hereditary tendency in this family toward some sort of abnormal development of the hands or feet.

transmissible change in the character of an offspring from that of his parents,—in other words, a sport. An early theory which was widely held was that of annular grooves or amniotic adhesions during intra-uterine life. However this, as well as the neurogenic theory, is no longer acceptable. Although time will not permit a detailed discussion of the subject, the weight of experimental and genealogical surveys leads to the conclusion that genetic factors are the principal causes of anomalies of the hand. One can impute imperfect development to factors residing in the genes—that is, to the germ plasm itself—or these factors may be transient, as for example poor nutrition, toxic influences, and the like. These latter might affect the embryo or foetus itself, but not the germ plasm of the offspring; thus the disturbance would appear in one generation but not in subsequent generations. It is well known that a pregnant mother who suffers from measles during the early weeks of pregnancy runs a very high risk of bearing a child with congenital defects, such as cataracts, heart disease, deaf-mutism, or mental deficiency. Nutritional deficiencies have also been shown to affect the offspring.

Stockard, in classic experiments with Fundulus and other fish, altered the normal rate of development by lowering the temperature or reducing the oxygen supply at a critical time in embryonic development, and thereby produced a series of monsters and twins. He concluded that the type of abnormality depended primarily on the stage at which the interruption in normal development occurred rather than the specific disturbing agent.

Experimental irradiation of the uterus and ovaries has also been shown to produce abnormal offspring. Bagg exposed mice to roentgen rays both prior to pregnancy and at various stages in foetal development. Offspring *in utero* showed profound arrests in

development, with the eye and brain especially affected. Many defects were those of the limbs, the most common being club-foot with dorsal flexion and syndactylism. Hypodactylism, congenital amputation, and polydactylism were also found, as well as blindness, other eye defects, and defects or absence of one or both kidneys. These abnormalities, which were studied for as long as nineteen generations, were often clearly inherited as Mendelian recessives. Bagg, confirming Stockard's hypotheses, expressed the opinion that the disturbing factor itself is of less importance than the time at which the disturbance occurs. Very early irradiation produced eye defects; later, those of the brain and branchial system; and still later, of the viscera. Bagg's work could explain the co-existence of multiple anomalies, such as those of eye, limb, and kidneys. The cogent point of these experiments is that the germ plasm had been altered; therefore, the anomalies were inherited subsequently.

A number of investigators have made observations on the effects of irradiating the pregnant woman, and there is evidence to support the view that irradiation may produce several kinds of anomalies, including those of the extremities.

There can be no doubt that many hand anomalies are inherited; this assumption has been substantiated by numerous authentic and carefully gathered case histories (Fig. 1). Many geneticists claim that the anomalies are inherited as Mendelian dominants, and yet the trait may skip one generation, may alter its form of expression, or may otherwise behave in an atypical manner. To explain these failures of expected ratios, the geneticists use many explanatory and qualifying terms such as "variable expression", "variations in penetrance", "inhibiting or modifying genes", "environmental factors", or "incomplete dominance".

Harris, from a study of human embryonic material, estimated that 5 per cent. of patients showing deformities of the extremities had a family history of abnormality. Murphy found a family history of defects in 7 per cent. of his cases. Our own series, though admittedly small for statistical interpretation, also showed indications of hereditary factors in 5 per cent. of the cases. The small percentage of cases with family history may be due to one of two reasons: First, it is quite possible that parents deliberately conceal a family history, feeling that in some way this reflects on themselves, or they may be truly ignorant of the occurrence of the character in other members of the family; second, this may be the first appearance of a trait that is later seen to be a genic mutation.

Birch-Jensen, in discussing the eugenics of hand anomalies, goes so far as to advise against marriage of persons with certain malformations; in other instances he mentions the possibility of induced abortion or even sterilization. Theoretically this might accomplish a reduction of the frequency of hand anomalies, but centuries or even millennia might be needed. If carriers of a defective gene have the expected life spans and produce the normal number of offspring, ". . . the proportion of the defective genes in the 'pool' of the sex-cells of the population would remain constant. . . . The gene proportions remain constant also if equal numbers of the genes are pumped out of the 'pool' by death and pumped into the 'pool' by mutation", as stated by Dunn and Dobzhansky.

We have not found a clinical classification of hand anomalies which seems adequate, nor do we offer one here, for we feel that it is impossible to do so. Many congenital anomalies of the hand are not simple defects but are complicated, combining a number of related or unrelated malformations. In some instances it is difficult even to decide the main category in which certain cases belong. For this reason we have not tabulated the results in our series of cases.

Since this study is based primarily upon our own cases, it is not all-inclusive and there are a number of anomalies that will not be considered.

SYNDACTYLISM

Syndactylism appears to be the most common congenital deformity of the hand.

FIG. 2-A FIG. 2-B

Fig. 2-A: Simple syndactylism involving the middle finger and ring finger. The web was so close that an attempt to cover the areas by flaps from the fingers, after they had been separated, would certainly have been unsuccessful.

Fig. 2-B: Postoperative photograph shows that the nail matrices on each side of the split had been ablated in order to bring the free skin graft around the nail margins.

According to Bunnell, it occurs "once in every one to three thousand births". The condition has been described in gibbons, various marsupials, and some other mammals. Webbing in humans is most frequent between the middle and ring fingers and between the second and third toes; there are some indications that the condition is almost twice as frequent

FIG. 3-A FIG. 3-B FIG. 3-C

Fig. 3-A: Ectrosyndactylism involving four fingers and the thumb. The web of the thumb extended along its entire length.

Fig. 3-B: Photograph taken after the thumb web had been split and a free skin graft applied. Since the use of the hand was greatly restricted, it was felt that the development was being hindered. The thumb web was, therefore, split and grafted when the child was fourteen months old.

Fig. 3-C: Preoperative roentgenogram showing the disparity in size of the metacarpals, the second being the largest and the third the smallest.

in males as in females, although these data have been questioned. Syndactylism is often associated with polydactylism, and in some families a single gene may cause both anomalies. The condition may also be associated with hypodactylism and deficiencies of the long bones.

Warkany and Nelson, in 1941, showed that offspring of rats reared on deficient diets showed many abnormalities: shortening or absence of the tibia, fibula, radius, ulna, and bones of the hands and feet with syndactylism. In addition to these abnormalities of the extremities, there were also shortening of the mandible, fusion of the ribs, and deformities of the sternum, maxilla, clavicle, and scapula. Recent studies indicate that the essential substance missing from the diets was riboflavin.

In our series we had eighteen cases of syndactylism, six bilateral and twelve unilateral. Of this total number, six cases of simple webbing involved only two fingers, in each instance the third and fourth digits. Of the whole group, there were seven patients with short fingers in addition to the syndactylism. These were not cases of "uterine amputation", but were true short fingers.

In addition to this group, eleven other patients had syndactylism as well as another congenital anomaly (not counting claw-hand).

FIG. 4-A FIG. 4-B

Preoperative and postoperative views of another type of syndactylism in a two-year-old girl (member of the "X" family). The patient's left hand was normal.

FIG. 4-C FIG. 4-D

Fig. 4-C: In the preoperative roentgenogram an extra bone, resembling a phalanx, lay transversely between the metacarpophalangeal joints. Note the deviation of the ring finger.
Fig. 4-D: Postoperative roentgenogram.

FIG. 5

Photographs of mother of patient shown in Figs. 4-A, 4-B, 4-C, and 4-D. Her left hand had been operated upon elsewhere in childhood. Her right hand had not been operated upon.

Thus twenty-nine, or almost half of the entire group of sixty-two cases, had syndactylism, either alone or in combination with another defect.

The most common type, simple skin webbing of the third and fourth digits, is also the mildest form (Figs. 2-A and 2-B). Syndactylism may, of course, involve all the fingers, which sometimes present a complete "mitten" effect. We have only one case in which the thumb and four fingers were involved. The condition not only varies in the number of digits involved, but also in degree (Figs. 3-A, 3-B, and 3-C). Thus, in some cases there is a shallow webbing, while in others the web may extend the entire length of the joined digits. Far more serious involvements occur when the bones are fused; sometimes even the tendons and nerves are joined, with a double nail at the end of the finger (Figs. 4-A, 4-B, 4-C, 4-D, and 5).

Needless to say, one should have careful roentgenographic studies to determine the

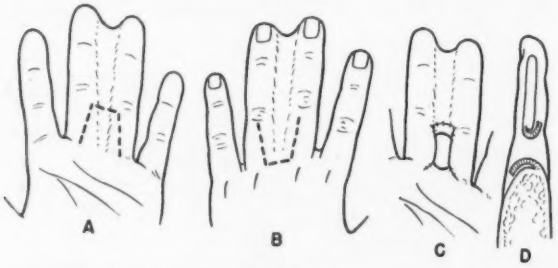

FIG. 6

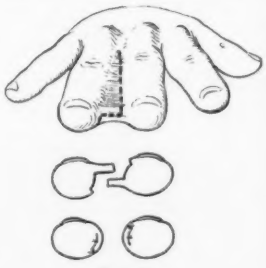

FIG. 7

Fig. 6: Félizet's operation (1892) introduced a new and interesting principle. Palmar and dorsal flaps were utilized as shown in *A* and *B*. They were reflected, one at the base of the digital fold and the other distally, and the wound edges were sutured as shown in *C*. In the original operation, at a second stage, the remainder of the web was severed. This operation was an improvement on Zeller's, but employed the same principle,—that of a flap across the commissure.

Fig. 7: Didot's operation for syndactylism. This type of operation is of historical rather than practical interest, for the web is rarely wide enough to permit its successful use. An unsuccessful result with this type of operation jeopardizes any future repair. (Reprinted by permission from *Principles and Practice of Plastic Surgery*, by A. J. Barsky. Baltimore, Williams and Wilkins Co., 1950.)

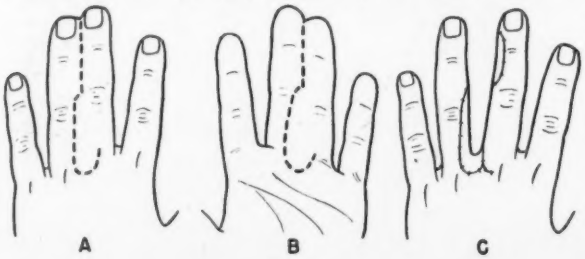

FIG. 8

Radulesco (1923) used flaps of a slightly different design in an effort to obtain sufficient tissue to wrap around the fingers. Similar types of flaps are still reported from time to time. None of them can provide sufficient skin to cover *all* of the denuded areas. *A* shows the flap from the dorsal surface and *B*, from the palmar surface; *C* illustrates how a free graft may be used to supplement the flaps.

condition of the bones, and the greatest care must be taken during operation to avoid damaging tendons, nerves, or the vascular system, which may be fused or otherwise involved.

Theoretically there are two main types of operative procedure for the repair of syndactylism,—a flap operation, a free skin graft, or a combination of the two. Didot's classic operation is of the flap type; however, since the web is rarely, if ever, wide enough for this type of operation, it is strongly condemned. An unsuccessful operation of this nature will result in the production of scar tissue and will not only make a secondary repair more difficult, but will also decrease the chances for a functionally perfect result. It is interesting to note that as late as 1940 the Didot flap type of operation was still

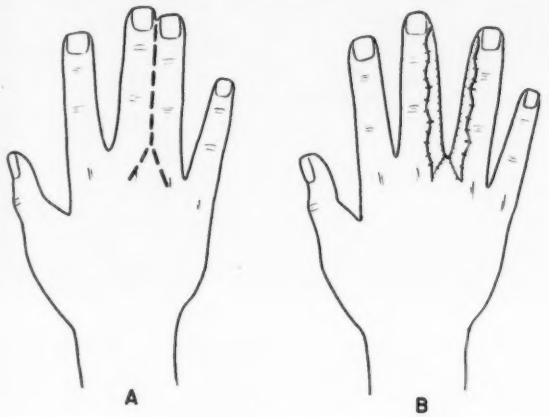

FIG. 9

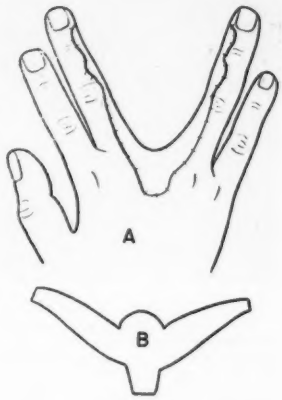

FIG. 10

Fig. 9: The most successful type of procedure is one in which an inverted V-shaped flap is taken from either the dorsum or palm of the hand, or both. After the fingers have been separated, the flap or flaps are spread across the commissure and the remaining areas on the lateral aspects of the fingers are covered with a free full-thickness skin graft. The result will be more satisfactory if the incisions are curved instead of straight. The use of the flap was first suggested by Zeller in 1810.

Fig. 10: Frequently a case is encountered in which a flap type of operation has been tried unsuccessfully. The resulting scar tissue makes it impossible to use the flap across the commissure. In this type of case, only a free skin graft should be used. This should be generous in size and should extend well onto the dorsal and palmar surfaces, to prevent a later "creeping up" of the commissure.

FIG. 11-A

FIG. 11-B

FIG. 11-C

Fig. 11-A: Syndactylism with annular grooving. The thumb and the index finger were normal. The remaining three digits were webbed and had annular constricting grooves. Note the dimples at the bases of the webs.

Figs. 11-B and 11-C: Postoperative result. The bands have been relieved and the fingers have been separated and grafted.

being advocated (Figs. 6, 7, and 8). A better operation consists in dividing the web and skin-grafting the raw areas thus produced. Should bone fusion be present, it is of course necessary to separate the bone of the fused digits. Early operation on infants and young children should be performed only if it is felt that the development of the fingers may be retarded by the condition. A better surgical result will usually be obtained if the operation

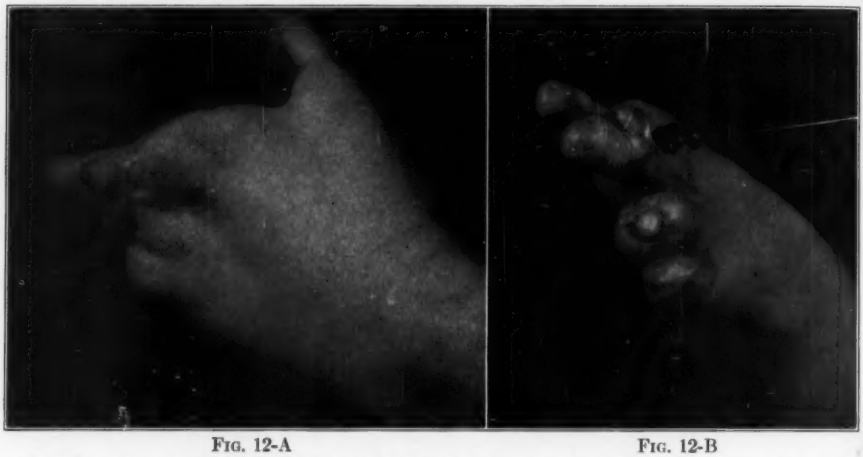

FIG. 12-A FIG. 12-B

Fig. 12-A: Syndactylism with partial absence of the fingers, and annular grooves. Note the dimple on the dorsum of the hand. This corresponds to the base of the web and often consists of a tunnel with epithelial remnants, which must be dissected away carefully. It is not wise to separate and graft all the digits at one time, because of the possibility that the central fingers may not have enough blood supply.

Fig. 12-B: The stumps of the ring finger and little finger have been separated and grafted. A probe is inserted into the epithelial-lined tunnel, referred to in Fig. 12-A.

FIG. 12-C FIG. 12-D

Fig. 12-C: All four finger stumps have been grafted. This patient had a very useful hand and remarkable dexterity. She played the piano and was employed as a secretary and typist.

Fig. 12-D: The patient is wearing a glove-type "dress" prosthesis on the left hand. The right hand showed a very mild annular groove at the base of the index finger. This was the only defect in the hand and did not interfere with function.

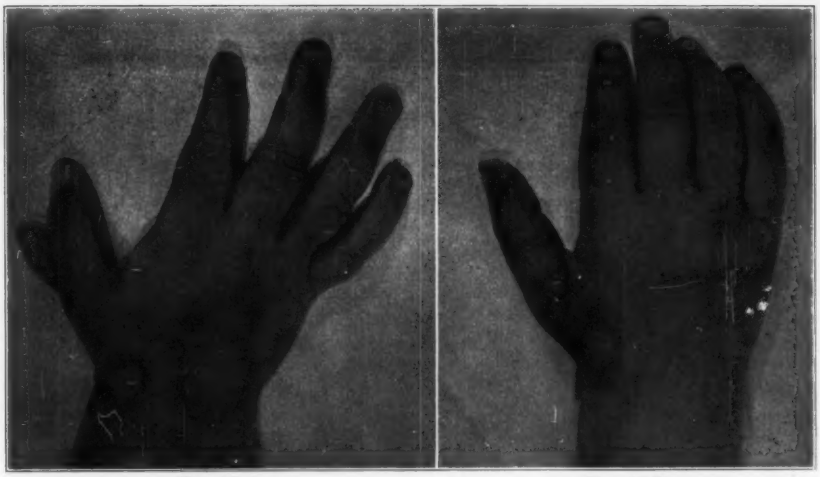

FIG. 13-A FIG. 13-B

Fig. 13-A: Simple polydactylism, with the supernumerary digit located on the radial side of the thumb; its removal is a simple procedure. If the capsule of the joint is involved, it should be closed at operation. Any tendons attached to an accessory digit should be transferred to the adjacent digit.
Fig. 13-B: Postoperative view of hand.

can be delayed until the child is at least two years, or preferably even older. The normal position of an infant's fingers is in flexion, and immobilization of these very small digits is often extremely difficult. However, one should not maintain too rigid an attitude in so far as the age of operation is concerned.

The operation of choice, if the web permits, is to take a small V-shaped or U-shaped flap from the dorsum of the finger, pedicle proximal, and to reflect this across the base of the web space after the fingers have been separated (Fig. 9). The remaining denuded area should be covered with a free skin graft, either three-quarters or full thickness. In the event that it is not possible to use a small flap to cover the base of the web space, it is well to pattern the free skin graft so that small rounded or pointed projections from the skin graft may be drawn dorsally and volarward when the graft is sutured (Fig. 10). A pressure bandage and a well padded splint should be applied.

If a double nail is present, it should be split, and a portion of the nail and its matrix on each side of the split should be ablated so that the graft may be brought around the nail margin. If more than two fingers are fused, one should be very cautious about separating both sides of a digit; it is best to do one side at a time, in order to avoid possible damage to the circulation. In our cases we have not encountered one in which there has been a common tendon for two fingers. However, should this be the case, the ten-

FIG. 14

Polydactylism. This patient, who came for treatment of a hand wound, refused to permit removal of the extra thumb.

don should not be split, but should be left attached to the better finger. Later a tendon graft may possibly be placed in the other finger (Figs. 11-A to 11-C and 12-A to 12-D).

SYMPHALANGISM

Symphalangism, a fusion of the interphalangeal joints, usually of the middle phalanx with the proximal or distal, occasionally accompanies syndactylism. I have not found it

FIG. 15-A

FIG. 15-B

Fig. 15-A: Polydactylism, with the supernumerary digit on the ulnar side of the hand, between the ring finger and little finger.

Fig. 15-B: Postoperative result.

Fig. 15-C: Preoperative roentgenogram of this hand.

FIG. 15-C

in simple cases of syndactylism, but only in the more complicated types, those in which there were extra phalanges present or in which brachydactylism was found.

Symphalangism, like many other congenital malformations, is inheritable. There is not much that can be done for the condition, if found in distal and middle interphalangeal joints, aside from arthrodesing the joint in a more useful position of partial flexion. Although arthroplasty has been advised, it is well known that this type of operation is satisfactory in the metacarpophalangeal joint, but not too successful in the middle and distal finger joints.

POLYDACTYLISM

Polydactylism is a part of the general phenomenon of duplication of parts. Examples range from complete duplication of a limb to doubling of a single phalanx. The condition is found not only in man and monkeys, but in horses, cattle, dogs, cats, guinea pigs, mice,

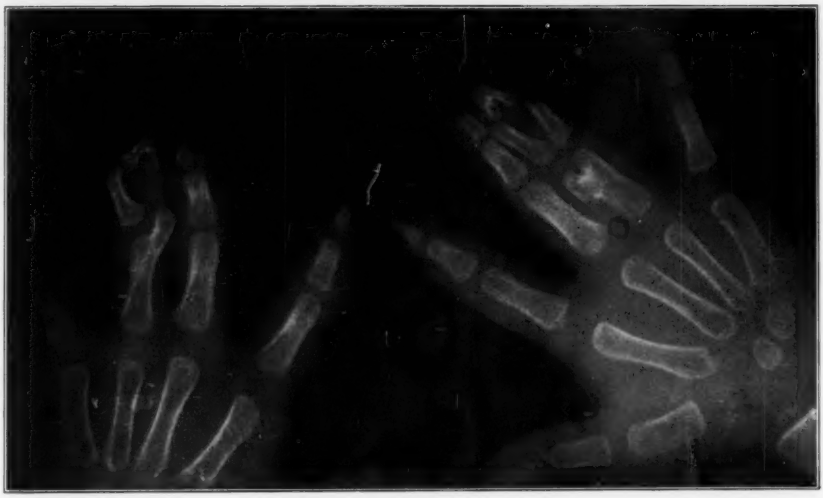

FIG. 16

Roentgenograms of bifid ring finger, bilaterally. The patient's mother had a similar defect. (These were members of the "X" family indicated in Fig. 1.)

fowl, and other animals. Polydactylism tends to be inherited, and not uncommonly accompanies syndactylism, brachydactylism, brachyphalangism, or other congenital anomalies. There is a tendency for the condition to be marginal—that is, the accessory digit is present on either the ulnar or radial side of the hand—rather than to be centrally located. In hexadactylism, which is the usual form, according to Müller radial duplication of the

FIG. 17-A FIG. 17-B

Figs. 17-A and 17-B: Photograph and roentgenogram of a double thumb.

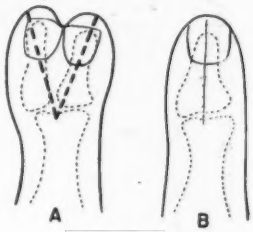

FIG. 18-A

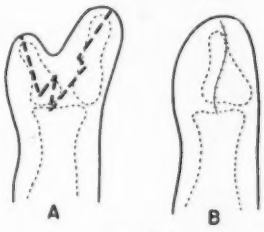

FIG. 18-B

Fig. 18-A: Bilhaut-Cloquet operation for bifid thumb. A bifid digit may be regarded as a form of polydactylism in which the duplication has not proceeded beyond the terminal phalanx. This type of duplication is occasionally seen in the other fingers, and may be corrected by the same method. When both parts of the bifid thumb are of about equal size a central wedge-shaped excision, as shown in *A*, should be carried out. The closure is shown in *B*.

Fig. 18-B: In the Bilhaut-Cloquet operation, if one part of the bifid thumb is larger than the other, a zigzag incision will be required. This is made on the flexor surface of the thumb, as shown in *A*. When the short component is stretched, the two sides may be made more nearly even.

thumb is more frequent than ulnar duplication,—that is, an extra little finger. Radial and ulnar polydactylism may occur together in the same family, although they are probably determined by separate genes.

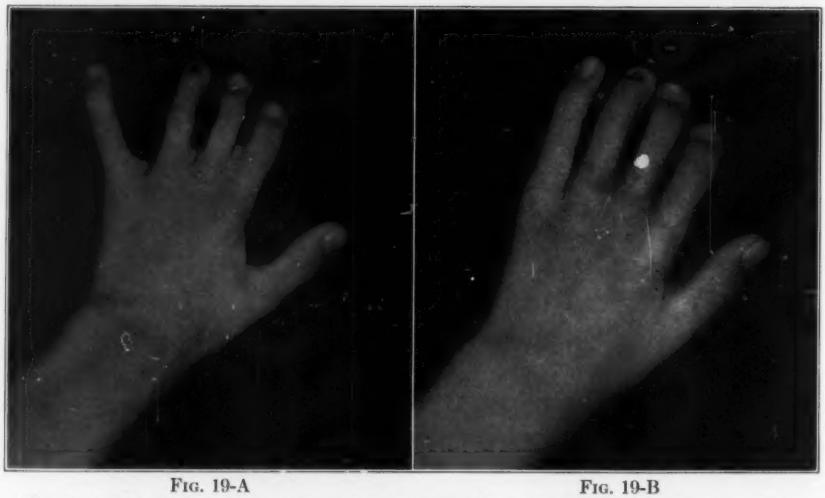

FIG. 19-A FIG. 19-B

Fig. 19-A: Brachydactylism accompanied by moderate syndactylism in a nine-year-old boy. Note the deformed nail on the ring finger. The mother stated that this was congenital.

Fig. 19-B: Postoperative result. The procedure followed is illustrated in Fig. 9.

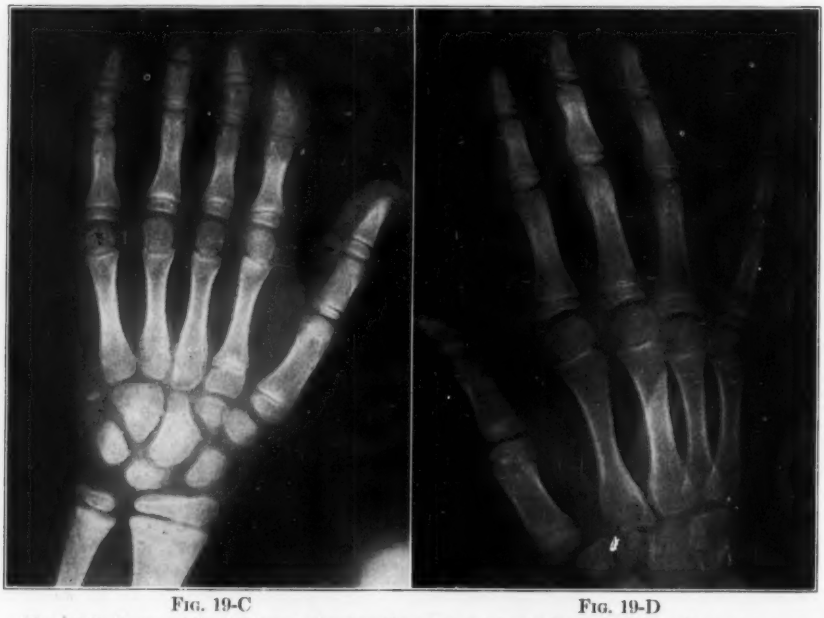

FIG. 19-C FIG. 19-D

Fig. 19-C: X-ray of the left hand, showing the extra epiphysis, located proximally in the second metacarpal, and the disproportion in the lengths of the phalanges. In this type of case the finger joints are not affected and the profundus tendon is attached to the distal phalanx; therefore, the patient can flex the distal joint. There is a suggestion of epiphyses at the distal ends of the proximal phalanges of the index finger, middle finger, and ring finger. The entire left hand was smaller than the mate, as can be readily seen by comparing with Fig. 19-D.

Fig. 19-D: X-ray of normal right hand of patient.

Embryologically, in man, digitation is well marked by the seventh week of foetal life and is completed by the eighth week. Lineback reported unilateral polydactylism in a 22-millimeter foetus.

The anomaly occurs in three main types. In the first, there is an extra fleshy mass, not adherent to the skeleton and often without bones or cartilage, muscle or tendons. In the second, there is duplication of the digit, which is more or less like an ordinary finger or toe, contains bones, and is articulated with an enlarged or bifurcated metacarpal. The third type, which is quite rare, is that of an extra digit with its own metacarpal or metatarsal bone. All possible intergrades occur between these forms (Figs. 13-A to 15-C).

In our own series there were eleven cases, nine unilateral and two bilateral. Eight had the extra digit radially located, including a bifid thumb. Three patients had the extra digit on the ulnar side; of these three, two were bilateral. In our cases we have not seen more than one extra digit on each hand. However, one patient had seven toes on one side and six on the other. Beck cited a case of Voigt's with thirteen fingers on each hand and twelve toes on each foot.

Careful study is required to decide in some instances not only the advisability of operation, but which digit should be retained. In the case of the

FIG. 20-A

Brachysyndactylism of the right hand.

FIG. 20-B

Roentgenograms showing the rudimentary middle phalanges, especially of the index, middle, and ring fingers. The webbing of these fingers can be seen in the soft-tissue shadows. This patient had flexion only at the metacarpophalangeal joints and one finger joint.

thumb, one should be extremely vigilant to see that tendinous attachments are preserved, as well as the attachments of the intrinsic muscles. In many instances the decision and operation are simple, but with the thumb, transfer of a tendon or muscle from the accessory digit may be necessary.

We can regard the bifid thumb as a form of polydactylism,—one in which the duplicating process did not proceed beyond the terminal phalanx. Although duplication of the distal phalanx is said to occur most frequently in the thumb, we have found it in two cases in the ring finger (Figs. 16, 17-A, and 17-B). In a bifid thumb, excision of a central wedge

FIG. 21-A FIG. 21-B

Fig. 21-A: In another type of brachysyndactylism, the webbing was more pronounced on the ulnar side of the hand. The rudimentary fingers were thin and weak.
Fig. 21-B: Postoperatively, the four fingers were of approximately the same length.

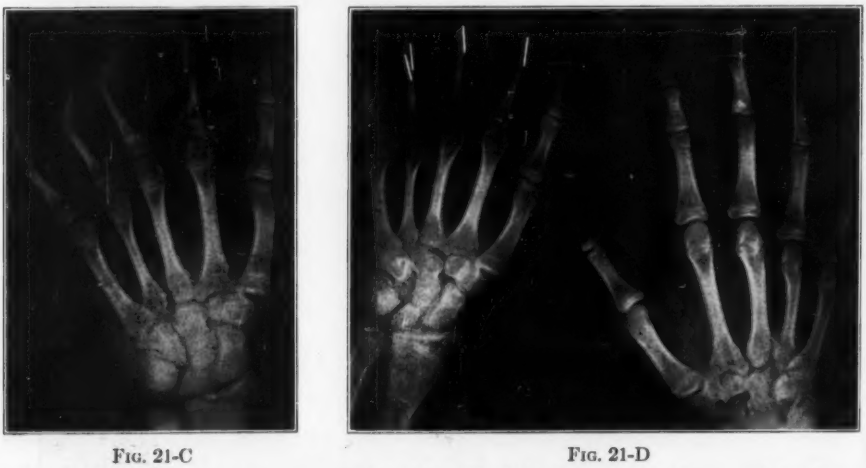

FIG. 21-C FIG. 21-D

Fig. 21-C: Preoperative roentgenogram of the hand. The index finger, middle finger, and ring finger had only two phalanges; the little finger had three, including a rudimentary middle phalanx. There were no muscle attachments on the distal phalanges of the index, middle, and ring fingers. These were flail joints.
Fig. 21-D: The interphalangeal joints of the index, middle, and ring fingers have been arthrodesed and the Kirschner wires are still in place. The arthrodesis was carried out after the fingers had been separated and grafted. Note the disparity in size between the two hands.

Fig. 22-A: Annular constricting grooves visible on the thumb, index finger, middle finger, and ring finger.

Fig. 22-B: Postoperative photograph. The annular grooves were excised in two stages; half the circumference was treated on the proximal band and the opposite half on the distal band. It is not wise to encircle the finger completely at a given place.

The operation should be performed under pneumatic tourniquet, since the digital nerves in these small hands are easily damaged.

FIG. 22-A FIG. 22-B

from each tip and joining the resulting segments (Bilhaut-Cloquet operation) is the operation of choice (Figs. 18-A and 18-B).

BRACHYDACTYLISM

Brachydactylism was the first example of Mendelian inheritance demonstrated in man, and is said to be a simple Mendelian dominant. The embryonic development of the fingers may be interfered with so that one or even all of the digits are shortened. This can result from decreased length of the phalanges, or decrease in the number of phalanges, or sometimes to a shortened metacarpal. The condition occurs frequently in conjunction with webbed fingers or with polydactylism (Figs. 19-A to 19-D, 20-A, 20-B, and 21-A to 21-D).

In some instances the fingers function well,—that is, the profundus tendon is attached to the distal phalanx and is able to flex the fingertip. In other cases there appear to be no muscle attachments to the terminal phalanges, and the distal interphalangeal joint is flail, so that the patient is required to back up the distal phalanx with the remaining fingers in order to make a pinching motion. Still other patients have a rigid joint which does not function.

FIG. 23 FIG. 24

Fig. 23: Absence of the fingers, together with a constricting band and rudimentary appendage on the ulnar side of the hand.

Fig. 24: Congenital absence of the thumb.

FIG. 25

Absence of fingers. On the left hand the patient had a thumb and three fingers, on the right hand a thumb and two fingers; at the wrist on the ulnar side was a rudimentary appendage with no muscle attachments.

In our series we had seven cases of short fingers, and seven additional cases of brachydactylism in conjunction with annular grooves and syndactylism. In the first group each digit is separate, though there may be a slight degree of webbing of the proximal phalanx.

If brachydactylism occurs with syndactylism, the web may be repaired, as already outlined. If there is a flail joint, this may be arthrodesed at the proper time so that the patient has a stable finger tip. No treatment is necessary for symphalangism.

ANNULAR GROOVES AND CONGENITAL AMPUTATIONS

The condition may be simply a very shallow groove in the skin or subcutaneous tissue, involving only one digit, or there may be multiple deep grooves extending down almost to the bone, with a small distal segment of the finger attached by a pedicle. Annular grooving also occurs in the foot, leg, and forearm. Although we have seen a number of cases involving the leg, foot, and the fingers, the author has never seen a case in which the forearm was involved.

We have had seven cases of annular grooving together with syndactylism and brachydactylism, but only one case in which this was the *only* defect of the hand. Another patient had a groove on one hand as its only defect, but the other hand showed syndactylism and partial absence of the distal portion of the phalanx.

A shallow ring, such as is seen in Figure 12-D, interferes in no way with the function of the finger. On the other hand, deeper rings or multiple rings are apt to cause oedema (Figs. 22-A and 22-B). The grooves should be excised down to normal structures, the

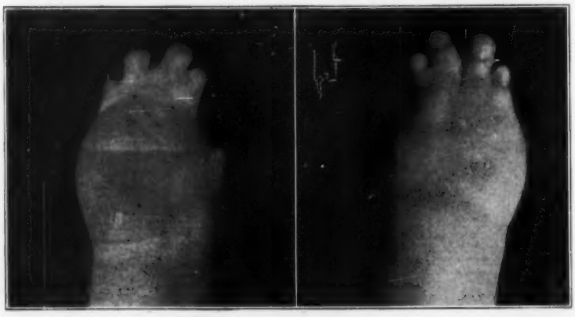

FIG. 26-A FIG. 26-B

Fig. 26-A: Palmar view of the right hand of a two-year-old boy, showing absence of the thumb and ectrosyndactylism.

Fig. 26-B: Dorsal view.

Fig. 26-C: Roentgenograms of both hands for comparison. Not only was the thumb missing, but the first, second, and third metacarpals were cartilaginous. The fourth and fifth metacarpals were rudimentary, the fifth being a good deal larger than the fourth. The four fingers were webbed, and there were only two phalanges in each. Only the proximal phalanx of the ring finger was seen to

FIG. 26-C

have an epiphysis. Note the disproportion in size between the abnormal and normal hands.

subcutaneous tissue properly approximated, and the skin scar zigzagged in order to avoid the possibility of an annular constricting scar. It is advisable not to encircle the finger completely at one operation, but to do half the finger at a time.

The literature pertaining to uterine amputations has run a wide gamut during the past century. Almost every possible explanation for the condition has been given at one time or another, including gangrene, inflammatory changes, diet, toxic factors such as measles, and the Rh factor. The theory which was most prominent and which was firmly held by many observers was that amniotic bands were the cause of uterine amputations (Fig. 23). Within the past two or three decades there has been a shift in emphasis toward genetic factors, and the theory of amniotic bands is now given little attention.

Streeter, in 1930, showed many cases of intra-uterine amputation wherein "sharply circumscribed areas of limb-bud tissue are of such inferior quality that only imperfect histogenesis occurs". These finally slough off in development and are said to be amputated, not because of amniotic bands or other agencies external to the embryo, as was formerly

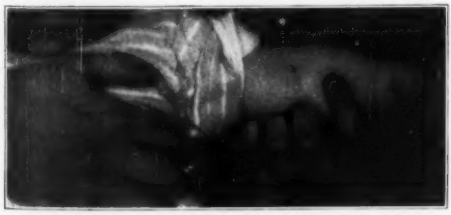

FIG. 27-A

Fig. 27-A: Absence of the left hand.

Fig. 27-B: Roentgenograms of both hands, showing complete absence of the carpals, metacarpals, and phalanges on the left.

FIG. 27-B

Fig. 28-A

Fig. 28-B

Fig. 28-A: Bilateral absence of the hand. Each side had one rudimentary digit which had no muscle attachments. This child also had congenital absence of the right foot.

Fig. 29-A

Fig. 29-B

Fig. 28-B: X-rays showing absence of the carpals and metacarpals. On one side there was a small bone resembling a phalanx, in the rudimentary digital appendage; there was none on the other side. This child was able to write and to feed himself, holding the utensils between his two stumps.

Figs. 29-A and 29-B: Roentgenograms of an eight-year-old boy whose hands were missing. On the right side (Fig. 29-A) there was a cleft between the radius and the ulna; a small rudimentary appendage was attached on the radial side. On the left side (Fig. 29-B) there was a rudimentary digit, also radially placed, containing three small phalanges. The carpals and metacarpals on both sides were missing. On the left side the patient had slight muscle control over the digit and was able to pronate and supinate the forearm.

supposed, but from genetic determinants. Streeter described sixteen cases of amputation of the digits and even larger portions of the extremities, and concluded that they are generally of germinal origin. At birth, only depressions, grooves, or healed stumps with residual strands may remain visible.

We had seven cases of complete absence of a digit, three bilateral and four unilateral (Figs. 24, 25, and 26-A to 26-C). Webster has emphasized the importance of conserving rudimentary digits and reattaching them to the hand.

We have had three cases of absence of the hand, two bilateral and one unilateral. In none of the cases was there a family history of any kind of hand anomaly. In the unilateral case, the child merely had a soft-tissue nubbin at the distal end of the forearm bones (Figs. 27-A and 27-B). In one bilateral case the lower two thirds of the leg and foot were missing on one side (Figs. 28-A and 28-B). This child had a very tiny nubbin of tissue on the radial side. He had no muscular control over this rudimentary appendage. Surprisingly enough, the child learned to write, holding the pencil between the two stumps. The third patient, an eight-year-old boy, on the left side had a rudimentary digit radially and very slight voluntary motion in the thumb; he was able to pronate and supinate the forearm. On the right side, there was a forklike separation between the radius and ulna (Figs. 29-A and 29-B).

FIG. 30-A

Fig. 30-A: Seven-year-old boy with congenital bilateral claw-hand and absence of the feet. In addition, the child was a deaf-mute.

Fig. 30-B: In the right hand there was a defect in the central ray, involving also the index finger. This type of cleft hand is usually associated with a foot defect.

Fig. 30-C: In the left hand there was a pronounced cleft, tapering proximally and dividing the hand into two parts; the digits on each side were webbed.

FIG. 30-B (above)
FIG. 30-C (below)

FIG. 30-D

FIG. 30-E

Fig. 30-D: X-ray of the right hand, showing the long proximal phalanx of the middle finger attached by soft tissue to the ring finger, which appeared otherwise normal. There was an extra bone between the heads of the second and third metacarpals. The index finger was irregular in shape and deviated, with twinning of the middle and distal phalanges.

Fig. 30-E: Roentgenogram of the left hand, showing the fourth and fifth metacarpals to be fused, as were also the first and second metacarpals. The patient was able to grasp by adducting the claws.

FIG. 31-A

Fig. 31-A: Fifty-year-old Puerto Rican woman with bilateral claw-hand and claw-foot.

FIG. 31-B

Fig. 31-B: Roentgenograms of the hands. In the left hand the middle finger was missing, but its metacarpal was present. The ring finger and little finger were webbed, as were the thumb and index finger. No finger contained more than two phalanges. In the right hand there were two phalanges for each digit. The ring finger and little finger were webbed in one group, and the three radial digits in another group.

Fig. 32-A: In another type of cleft hand, seen in an eight-week-old girl, the entire central portion of the left hand was missing,—that is, the metacarpals and phalanges. There was a metacarpal on the radial side and another on the ulnar side of the hand, each with two phalanges. A web extending from the radial digit, or thumb, to the other digit prevented abduction of the fingers. The right hand, shown for comparison, was normal.

Fig. 32-B: Postoperative result obtained by performing a Z-plasty on the web.

Fig. 32-C: The hand in use. These photographs were taken when the child was eight years old; she had surprisingly good function.

FIG. 32-A

FIG. 32-B

FIG. 32-C

CLEFT HAND OR LOBSTER-CLAW HAND

This is a rare congenital anomaly in which there is a defect of the central portion of the hand. The term is a rather loose one; our interpretation is that the individual has a

FIG. 33

Roentgenograms of a similar case. Both hands of a four-year-old child are shown for comparison.

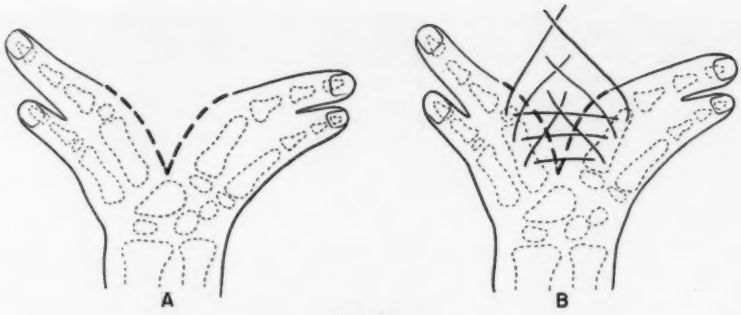

FIG. 34

The operation most often described for the type of claw-hand shown in Fig. 30-C. The margins of the cleft are freshened (*A*) and deep sutures are inserted to bring the metacarpals over, adjacent to each other (*B*). This was not done in the preceding case since it would have decreased, not improved, the function of the hand.

FIG. 35-A

Fig. 35-A: Congenital absence of the radius and club-hand in a four-year-old girl. Both hands were in extreme radial deviation.

Fig. 35-B: Showing normal carpals, metacarpals, and phalanges, with beginning signs of bowing of the ulna.

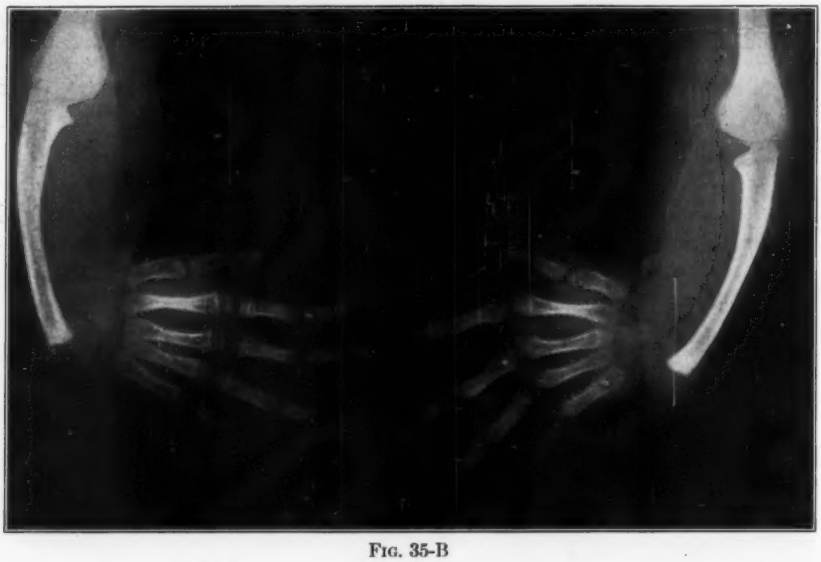

FIG. 35-B

defect of the central part of the hand and the remaining digits are divided into a radial and an ulnar group. In our own series we had four such cases,—two bilateral and two unilateral.

The consensus among investigators is that the condition is definitely hereditary. However, in our cases we were not able to secure any evidence of a hereditary tendency.

There are several varieties of cleft hand. The first type has a pronounced V-shaped cleft tapering proximally, dividing the hand into two parts; the digits in each part are generally webbed (Figs. 30-A to 30-E). The middle metacarpal as well as the finger is usually missing, and the hand resembles a true claw. The patient can often move the claws toward each other for grasping. In another variety of this same general type, the cleft does not extend so deep; the central metacarpal is present, but its digit is missing (Figs. 31-A and 31-B). Both of these types are usually bilateral and are associated with similar deformities of the feet.

There is still another type of cleft hand, consisting of a radial digit or thumb, with its metacarpal, and an ulnar digit (Figs. 32-A to 32-C). The remaining digits, together with their metacarpals, are missing. This type of cleft hand is generally unilateral and there is no associated foot deformity (Fig. 33).

FIG. 36

FIG. 37

Fig. 36: Roentgenogram of a two-year-old boy. Each upper extremity measured nine and one-half inches in length. The hand consisted of two fingers, each having a metacarpal and two phalanges. The fingers were webbed. The radius was present, but the ulna was missing. There was no movement at the elbow joint.

Fig. 37: Roentgenograms of the right and left sides of a five-year-old girl, exhibiting a rare anomaly of the hands and upper extremities. Both extremities were approximately the same size, but the x-ray plates were not reduced to the same size. The right humerus was approximately half its normal length, while the left was about three-quarters normal length. Both humeri were bifurcated at their distal ends. On the right side the medial bifurcation was much longer than the lateral portion; on the left side the bifurcations were more nearly even, although the lateral portion was larger than the medial. Arising from each lateral bifurcation was a rudimentary hand, consisting of two metacarpals and two digits, each with three phalanges. The patient was able to feed herself, but it was not possible to improve the function of the upper extremities. In addition, this child had bilateral knock-knee and marked curvature of the spine.

FIG. 38-A FIG. 38-B

Fig. 38-A: Bilateral megalodactylism in an eighteen-month-old boy. The middle finger and ring finger of the left hand were enlarged and were webbed.
Fig. 38-B: The right hand had a greatly enlarged thumb and index finger.

The treatment for lobster-claw hand varies with the individual case. For the first type, with the deep cleft, elimination of the cleft is generally advised, by the procedure shown in Figure 34. In our case (Fig. 30-C) we did not do this, since the patient was able to bring the two portions of the hand together in a pincer-like movement, and we felt that this function was more important than improving the appearance. We did separate and graft the webs. In the other type of web hand, with only an ulnar and a radial digit, the grasp of the hand can be improved by releasing the crest of the web through a Z procedure; after such an operation, children develop a surprising amount of dexterity.

DEFECTS OF THE FOREARM

Congenital absence of the radius in part or whole is fairly common; Kato collected 253 cases in the literature up to 1923, including three cases of his own. This condition is

FIG. 39

FIG. 40

Fig. 39: Eighteen-year-old male with enlarged fingers. The condition was caused by a cavernous hemangioma, although the patient stated it had been present at birth. This is not regarded as a case of true megalodactylism. The appearance was somewhat similar to that of enlargements which result from neurofibroma.
Fig. 40: Bilateral congenital absence of the long extensors of the thumb in a year-old male child. Operation on this child was deferred until a later time.

generally associated with club-hand. Absence of the radius is said to occur more frequently than absence of the ulna, and total absence is more common than partial absence. In 46 per cent. of Kato's cases the defect was bilateral; in 30 per cent., unilateral. Although any part of the radius may be missing, the presence of a remnant of the upper end is much more frequent than that of the lower. The radial bones of the carpus are either absent or fused; the soft parts of the radial side of the forearm may be affected. The ulna may be shortened, thickened, and curved; as the patient grows, the hand may be deviated radially until it is almost at a right angle to the long axis of the forearm.

We have had only one case of congenital absence of the radius. Figures 35-A and 35-B show this patient, aged four years, who has not yet been operated upon. Prior to any operation on the bones, it will be necessary to decrease the deformity by splinting and by operations upon the soft tissues, possibly lengthening the tendons of the index finger and thumb.

In general, it is necessary in this type of case to release the contracture, if any is present, and subsequently to arthrodese the wrist in a position of function. In 1928, Albee reported a successful procedure which he had tried in three cases. A tibial graft was mortised into the carpus below and into the shaft of the ulna above, to form a sort of strut. He believed it was not necessary to insert the upper end of the graft into the ulna, but simply to place it between the muscles on the radial side of the forearm. In addition, Albee corrected the bowing of the ulna by an osteotomy. He advised waiting until the age of three to operate.

Our second case of absence of a forearm bone (Fig. 36) occurred in a two-year-old child whose upper extremity measured nine and one-half inches in length. The ulna

FIG. 41-A FIG. 41-B

Fig. 41-A: Congenital bilateral anomaly. There were five fingers on each hand, but no thumb.
Fig. 41-B: X-rays of hands. The radial digits could not oppose.

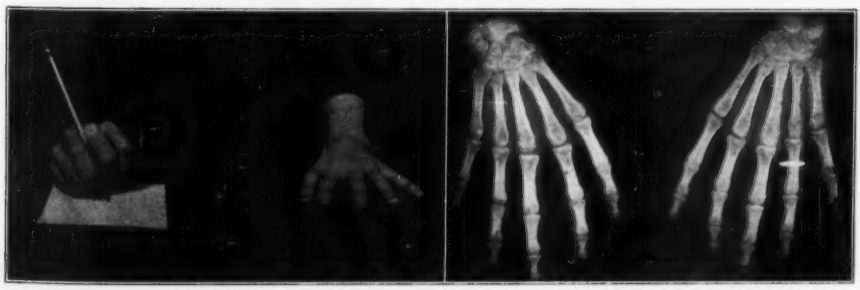

FIG. 42-A FIG. 42-B

Fig. 42-A: Photograph of hands of child's mother. She was able to carry on her duties as housewife, and felt that her activities were in no way hampered by the anomaly.
Fig. 42-B: X-rays of mother's hands. The radial digits could not oppose.

was missing; the radius was fused with the humerus. There were only two fingers on each upper extremity and two metacarpals. The fingers contained two phalanges and were webbed. In addition to the defects of the upper extremity, this child had congenital dislocation of the left hip and cryptorchism. He died while a subtrochanteric osteotomy was being done by an orthopaedic surgeon.

There are cases in which the arm or leg is absent, but there is a hand or foot present. The condition, which is referred to as phocomelus, is regarded as a defect in development at a certain stage, plus a possible hereditary factor. Few cases have been reported; in Hill's article published in 1937, he wrote that only three other cases had been reported since 1920. Among our series was one of this type.

MEGALODACTYLISM

Hypertrophy of the digits, called megalodactylism or macrodactylism, is a rare anomaly. The literature reveals that the second and third digits are most likely to be involved, and males more often than females. The process is local and not necessarily symmetrical. It is rarely hereditary, and the etiology has not been explained.

We have two cases of hypertrophy, in only one of which do we feel there was true hypertrophy,—that is, enlargement of all the elements of the finger. In one case an angioma was present at birth. The other, which we consider true megalodactylism, involved the thumb and index finger of the right hand (Figs. 38-A and 38-B); the middle and ring fingers of the left hand were not only greatly enlarged, but were fused as well. This patient was not operated upon, since his mother insisted upon a guarantee that the fingers would be completely normal after operation. In true megalodactylism, the only possible treatment is partial amputation, occasioned by the unsightliness of the condition.

If the condition is due to a neurofibroma or angioma (Fig. 39), removal of the excess tissue will improve the appearance of the finger. However, in neurofibroma the possibility of recurrence should not be overlooked. We also had one case of this type, but it is not listed in this series since it was not a congenital deformity.

MISCELLANEOUS

We had one rare case in which there was congenital absence of the long extensors of both thumbs (Fig. 40). Bunnell showed a picture of a similar condition; however, his case was unilateral, whereas our patient had bilateral involvement. This child will require a tendon transfer. Zadek also reported congenital absence of the extensor pollicis longus of each thumb. This he corrected by severing the extensor carpi radialis longus at its insertion into the second metacarpal; using a three-inch free graft from the extensor digitorum communis of the second finger, he thus extended and attached the extensor carpi radialis to the proximal part of the distal phalanx of the thumb. He made no attempt to form a tendon sheath. The operation gave thumbs that were almost completely functional.

We have two patients, a mother and son, with five fingers on each hand, but no thumb (Figs. 41-A, 41-B, 42-A, and 42-B). We are at a loss to know whether to classify these as missing digits or as polydactylism, since the patients have five fingers instead of four. The child has seven toes on one side and six on the other, and the mother has the same condition.

We have one case of unilateral deviation of the thumb, with no other defect; and two cases of flexion contracture of the thumb, one unilateral and one bilateral.

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THE USE OF A METALLIC FEMORAL HEAD *

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During the period May 1948 to December 1949, the author replaced the femoral head by a metallic prosthesis in ten hips. It is the purpose of this preliminary report to analyze these cases and to discuss some of the problems which have been encountered. The device, which first was used by Jaenichen in 1947, is known as the Jaenichen-Collison prosthesis. The author does not claim responsibility for its origin. Of the total cases in which it was used prior to 1950, his cases represent about 15 per cent.

Experimental work was conducted by Bohlman in 1929 on replacement of the femoral head, and since 1940 he has treated eight clinical cases, using a Vitallium device. In an additional case, Moore and Bohlman, in 1940, resected a large giant-cell tumor of the upper half of the femur and replaced the bone with a large Vitallium prosthesis. A good result was obtained, but the patient died twenty months later of a cardiac condition. The author's series is not concerned with such extensive replacement of bone, but only with replacement of the femoral head and neck.

Hudack and also McBride are known to have replaced the femoral head with plastic or metallic devices, but their results have not been published. Judet and Judet reported a series of cases of non-union, arthritis, and old dislocation, in which the femoral head was replaced by a device made of methyl methacrylate. This artificial head is made to fit snugly on the neck of the femur and has a long spike which passes down through the neck to the cortex. It requires sufficient bone to support the artificial head, a condition which often does not exist in cases of non-union and absorption of the neck.

ANALYSIS OF CASES

In the author's series, ten hips have been operated upon in eight patients; six cases were unilateral and two were bilateral. In five of the six unilateral cases there was non-union of the femoral neck; ankylosis due to rheumatoid arthritis was present in one unilateral and the two bilateral cases. It is necessary to consider each group of cases separately because of the marked differences in pathological findings and disability.

Of the five patients with non-union, there were four females and one male. Their ages ranged from fifty-five to eighty-two, with an average age of seventy-one years. The intervals since the original fracture varied from five to eighteen months, with an average of eleven months. None of the patients had undergone reconstructive surgery since the initial operation to reduce and internally fix the fracture at the time of injury; all had complete disability of the involved extremity and were unable to bear weight. In four a Smith-Petersen nail had been employed and in one a Lloyd-Collison screw. There was overriding of from one-half to one and one-half inches with an average shortening of one inch.

In the three cases of arthritis, representing five of the hips in this series, the disease was of the Marie-Strümpell type. The patients' ages were thirty-two, thirty-three, and forty-four, with an average age of thirty-six years. The average duration of the ankylosis was three years, but the duration of the disease process was much greater. The two bilateral cases occurred in males; the unilateral case occurred in a female. Both of the men had severe involvement of the spine with marked flexion deformity, and one had a fracture of the neck of the left femur with non-union. The woman with arthritis had a straight spine with marked limitation of motion; arthrodesis of the opposite hip had been performed elsewhere because of an erroneous diagnosis of tuberculosis. Her remaining hip had subsequently become involved and Vitallium-cup arthroplasty had been performed;

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this hip was stiff and painful until the cup was removed and the femoral head was replaced with a metallic head, in November 1949. This is the most recent case in the series.

One other patient in this group had undergone Vitallium-cup arthroplasty (Fig. 2-B), performed on the right hip in May 1948. The left hip had been fractured, non-union and deformity resulting, and was not considered suitable for cup arthroplasty. Therefore, the metallic femoral head was applied on May 28, 1948, as the first operation in this series (Fig. 2-C). For eight months he continued to use the Vitallium cup on the right and the metallic prosthesis on the left. At the end of that time, because of the better performance in the left hip, he requested that the cup be replaced by a metallic femoral head; this was done in January 1949.

It should not be inferred from these two instances that any conclusions are being made as to the relative merits of the Vitallium cup and the metallic femoral head. While other indications may include post-traumatic arthritis, hypertrophic arthritis, and deformities or tumors of the head and neck, the author's series has been limited to the Marie-Strümpell type of arthritis and to non-union due to fracture.

THE PROSTHESIS

The prosthesis, made of 18-8 SMo steel, consists of the head, neck, plate, and screws (Fig. 1). The total weight is about one and one-quarter pounds, but in no instance has the patient been aware of the weight. The head is a solid steel ball and is made in six sizes, each enlarged by one-eighth of an inch, from one and one-half to two and one-eighth inches in diameter. The neck is a separate unit of solid steel and is available in six lengths, with quarter-inch differences, varying from one-quarter of an inch to one and one-half

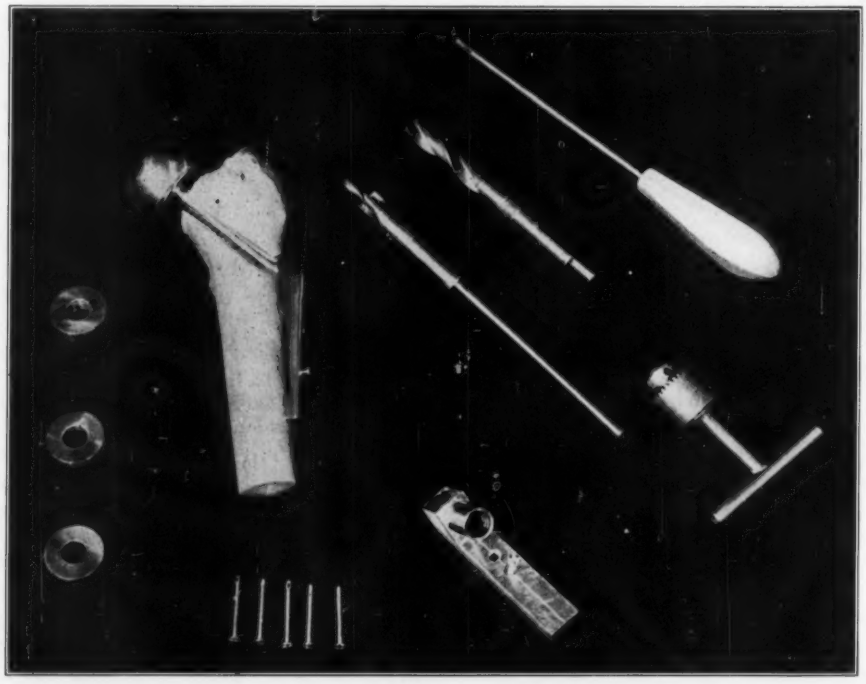

FIG. 1

The Jaenichen-Collison prosthesis consists of the head, neck, plate, and screws. Washers of varying thicknesses may be used to fill any defect remaining between the head and the bone. The special jig and drills are essential for accurate placement.

inches. One end of the neck fits snugly into a deep recess in the head and the other end into the angle of the plate. The upper end of the plate is angled to fit into the neck; the distal portion fits on the shaft of the femur. The plate is attached with the usual type of steel screw. The earlier plates required four screws; the more recent plates, six screws. A recent accessory for the prosthesis is a series of washers of varying thickness which can be inserted over the steel neck, between the head of the prosthesis and the bone, to provide support where the neck of the femur may have been absorbed or removed.

Reference should be made to the deficiencies which were encountered in the earlier models. No difficulty has arisen with the head or the neck portion of the device. The original plate was not sufficiently strong at the angle, and one plate in this series broke. It was replaced through a lateral incision without the joint being entered. The latest type of plate has been tested on a walking machine and has withstood a load of 250 pounds at the rate of thirty-six per minute for over two million simulated steps, without evidence of fatigue. In three patients, including the one in whom the plate had been replaced, the screws underwent fatigue fracture; the plate separated from the bone until the head rested on the remaining portion of the neck of the femur (Fig. 3-C). Subsequent improvement in the design of the plate provides for better distribution of the contact with the shaft of the femur and is intended to obviate the slight motion which apparently leads to fatigue fracture of the screws. Furthermore, six screws provide for better fixation than the original four. In the entire series of approximately 100 cases in which this device has been used to date, ten mechanical failures have been reported,—fatigue fracture of the plate in five cases and of screws in five.

Four of the failures have occurred in this series of ten hips, although the early equipment and technical assistance were identical in all of the cases. The mechanical failures occurred in unilateral cases in women above the age of sixty-four. In this category, sex and age should have been a protective influence, since these hips were subjected to much less stress than the five hips in the three male patients. The reasons for the failures, in addition to the structural weakness in the earlier models, include (1) lack of support to the metallic head due to insufficient bone to make contact; and (2) improper technique in the earlier operations, in which the device was inserted through a lateral incision. In the latter procedure, after removal of the head of the femur, the metallic head was inserted, the device was assembled in place, and the plate was brought against the shaft of the femur under considerable tension. Further reference will be made to these two features of the operation under discussion of technique.

TECHNIQUE

The patient is prepared in the usual manner for a reconstructive procedure on the hip. A long curved anterolateral incision is made along the anterior third of the crest of the ilium, thence downward from the anterior superior spine to below the muscle portion of the tensor fasciae latae, where it passes posteriorly across the lateral aspect of the thigh, sectioning the fascia lata for a distance of two or three inches (5 to 7.5 centimeters). The proximal two thirds of this incision represents the classical Smith-Petersen approach to the hip joint. The lower third permits the skin and tensor fasciae latae to drop back, exposing the vastus lateralis as far as the linea aspera, if desired. This exposure provides complete access to the hip and the upper third of the femur without the sacrifice of any nerve supply or muscle attachment. In case of non-union of the neck of the femur, the capsule is opened and the head is removed, but the acetabulum is not reamed. In case of ankylosis, a wider exposure is necessary with removal of the anterior portion of the joint capsule. Osteotomy is performed through the subcapital region of the neck, the head is removed, and the acetabulum is reamed to its original depth. Only enough of the neck is removed to accommodate the device to be inserted, so that sufficient bone is preserved to support the metallic head and to maintain the original length.

FIG. 2-A

Case 1. A. B., a man, aged thirty-three, had Marie-Strümpell arthritis with ankylosis of both hips and non-union following fracture of the neck of the left femur.

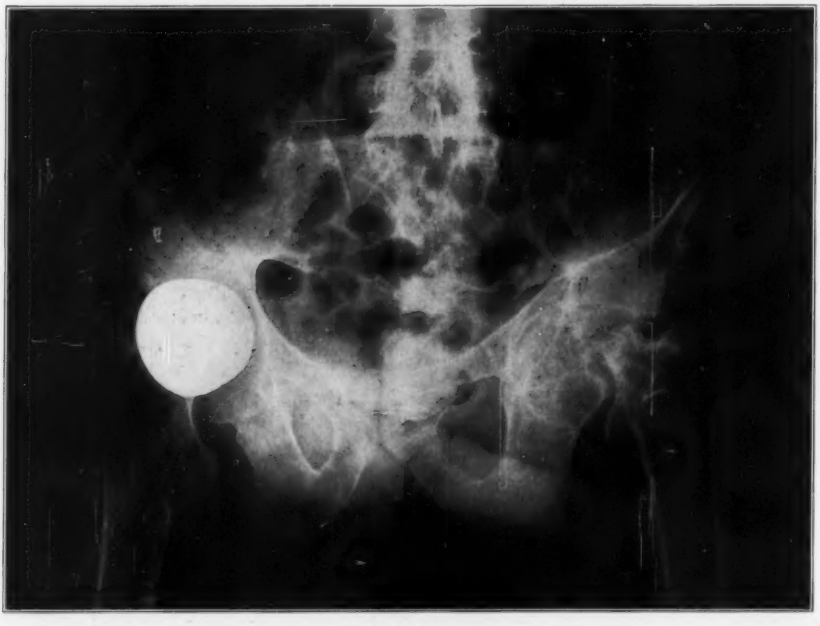

FIG. 2-B

Appearance after Vitallium-cup arthroplasty of right hip.

FIG. 2-D
Appearance sixteen months after replacement of the Vitallium cup.

FIG. 2-C
After insertion of the metallic femoral head on the left.

In this series, in only three instances was the acetabulum reamed; in the first case, this was somewhat deeper than necessary (Fig. 2-D). In the five cases of non-union and the two of replacement of the Vitallium cup, the acetabulum was not reamed. Where severe flexion deformity exists, as in arthritis, section of the tendinous portion of the iliopsoas is suggested; this was performed in three instances.

Following preparation of the acetabulum and the neck, the upper third of the shaft of the femur is exposed by splitting the posterior portion of the vastus lateralis. A long exposure is required, in order to insert the plate without excessive retraction. Although not used in this series, the alternative method of detaching the vastus lateralis at the linea aspera may be preferred. After exposure of the shaft of the femur, a special jig is attached with one screw. Through this jig the neck of the femur is drilled to the exact pattern of the prosthesis. The jig is then removed. The proper length of metal neck is determined and is placed in the plate before the latter is attached. This has proved easier than the earlier method of inserting the metal neck from the proximal end of the femur. The plate is then firmly clamped to the femur, and bottom and top screws are inserted. The appropriate size of metal head is applied, the acetabulum is given a final inspection, and reduction is accomplished. The remaining four screws are inserted and all screws are repeatedly tightened in turn so that the pressure on the plate is equally distributed. If the metal neck is too long, reduction may be difficult; if it is too short, the reduction is easily accomplished, but there remains a corresponding loss of length. Finally the motion of the hip is tested and the wound is closed in the usual manner.

POSTOPERATIVE MANAGEMENT

During the postoperative course there is a tendency for the extremity to externally rotate; if uncorrected, a permanent loss of internal rotation may result. Balanced suspension in a half-ring splint and Pearson attachment has proved a simple and comfortable

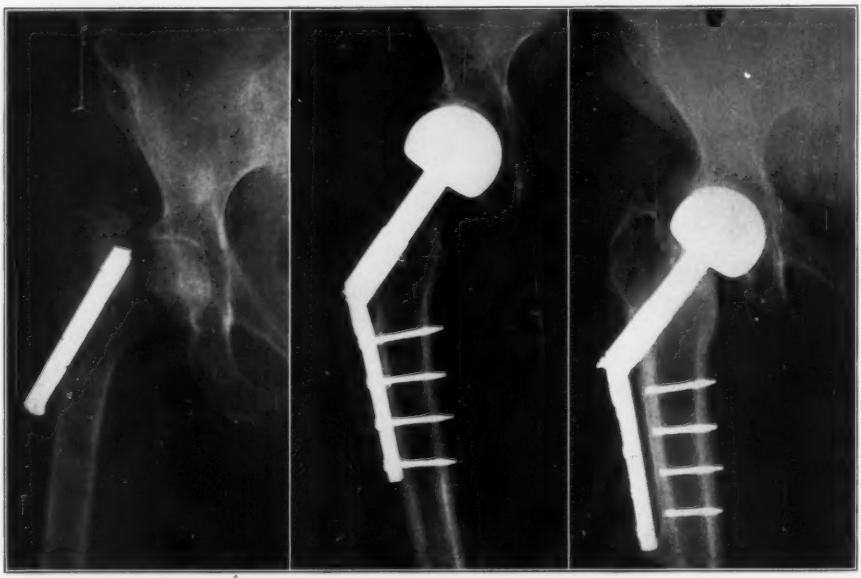

FIG. 3-A FIG. 3-B FIG. 3-C

Case 2. B. R., a woman, aged seventy-nine. Non-union of the femoral neck five months after internal fixation (Fig. 3-A), following insertion of the metallic femoral head (Fig. 3-B), and following fatigue fracture of the screws (Fig. 3-C). Note that the head has settled on the bone and that a bony shelf has formed beneath the plate. The patient is ambulatory without a cane.

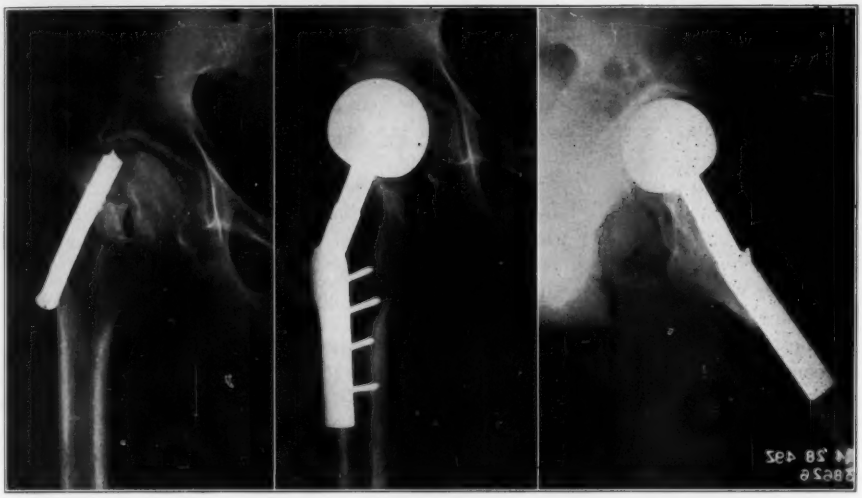

FIG. 4-A Fig. 4-B Fig. 4-C

Fig. 4-A: Case 7. J. B., a man, aged fifty-five. Non-union of the femoral neck, seventeen months after internal fixation.

Figs. 4-B and 4-C: Anteroposterior and lateral views after insertion of metallic femoral head. The patient has returned to his regular occupation as a sheet-metal worker.

method of immobilization. The extremity is kept in a neutral position for about a week; then support may be discontinued and the patient permitted up in a chair. Rigid immobilization is not required and early muscle exercise is encouraged,—first static contraction and then active motion of both the hip and knee. All the patients in this series put full body weight on the reconstructed extremity not later than the twenty-first postoperative day. In one case, unlimited weight-bearing was started on the sixteenth day. A period of three weeks' protection was chosen arbitrarily for these early cases, but there is no reason to delay weight-bearing after the period of wound healing. Since many of these patients have been bedridden for a number of months before operation, crutches are indicated until there has been sufficient return of general strength and particularly of quadriceps power. Except for intensive active muscle exercise, no special therapy is required, since the normal activities of sitting and walking will usually give the patient the maximum return of hip motion.

COMPLICATIONS AND SEQUELAE

One operative complication resulted in failure. A chronic infection occurred in the oldest patient, a woman, aged eighty-two, who had an uneventful postoperative course and began full weight-bearing at three weeks. Two weeks later she had increasing hip pain, and ten weeks after operation there was definite infection which persisted in spite of intensive antibiotic therapy. Ten months after operation the device was removed with osteotomy of the trochanteric region; this resulted in permanent healing of the infection. This patient is ambulatory and bears partial weight with crutches.

RESULTS

With the exception of the one failure due to infection, the other nine hips in this series are all in active use. The average follow-up period was eighteen months.

The two types of cases will be considered separately.

An analysis of the results in bilateral Marie-Strümpell arthritis is difficult because

of the extensive involvement of the spine. These cases are best evaluated by comparison of preoperative and postoperative disability and the degree of rehabilitation. The first patient with bilateral involvement was completely bedridden before operation (Figs. 2-A to 2-D). At this time, sixteen months after his second operation, he is pursuing a course in vocational training, can ride over 200 miles in one day by automobile, and drives his own car. Because of the marked flexion deformity of his spine, he uses two canes for walking, and does part of his travel by wheel chair.

The second patient with bilateral involvement had his last operation sixteen months ago. He had been ambulatory with great difficulty before operation. While he has gained considerable hip motion and is more comfortable when sitting and lying, he still requires crutches for walking, has slight flexion deformity of one knee, and associated quadriceps weakness. He is considered moderately improved.

The third patient, a woman with operative ankylosis of the opposite hip, underwent replacement of a Vitallium cup with the prosthesis, seven months ago. This is the only case in which free motion could not be demonstrated at the time of operation, after removal of the head of the femur. She has retained the limited motion then present, and has in fact gained slightly. While previously she could take only a few steps, she is now on her feet almost all day and is doing much of her housework.

The average range of motion in the five arthritic hips is as follows: flexion-extension 50 degrees, external rotation-internal rotation 50 degrees, abduction-adduction 30 degrees. These hips were almost completely ankylosed prior to operation. The nature of the disease does not yet permit final evaluation in these cases, but to date there has been no loss of motion since the early postoperative period. No mechanical failure has been encountered in the device in these five hips.

Of the five patients operated upon for non-union following fracture of the femoral neck, there was one failure due to infection; four are ambulatory with full weight-bearing. In all of these cases, discomfort is minimal. The patient in whom the plate and subsequently the screws broke, underwent the last operation two months ago; she resumed full weight-bearing twelve days later. The other two patients who have broken screws are walking,—one with the aid of a cane, the other without a cane (Figs. 3-A to 3-C). Their ages are seventy-nine and eighty-one years and, since their activities are not restricted, operation is not indicated merely to replace the defective screws. The fourth patient (Figs. 4-A to 4-C), who had been completely incapacitated for seventeen months before operation, returned to full work as a sheet-metal worker and roofer five months after operation and has continued daily in this capacity. The average range of motion in the four hips, including the two in which the screws are broken, is as follows: flexion-extension 100 degrees, internal rotation-external rotation 60 degrees, abduction-adduction 55 degrees. The average gain in length was one inch, so that the length of the extremity in each case was practically normal.

CONCLUSIONS

The steel femoral head has been well tolerated in the acetabulum and there has been no evidence of necrosis or protrusion except in the one case in which infection developed. Except in that case, there has been no occasion to remove the head of the prosthesis or to expose the new joint. Minimal discomfort has been present, and there has been no evidence of pain due to the metal. There has been no bone necrosis due to pressure or traction and no loosening of the screws. In one case with broken screws, a supporting ledge of bone has developed under the lower border of the plate and the patient is bearing full weight without a cane.

Structural defects have led to fatigue fracture of one plate and of all screws in three cases. This indicates that the device should be strong and that proper technique should be observed in its application. It is possible to replace the plate and screws through

a lateral approach to the femur without arthrotomy. A single long anterolateral incision is recommended, all muscle attachments being preserved. The head of the prosthesis or intervening washers should rest on bone if possible, although this is not mandatory if the device is sufficiently strong.

The minimum of postoperative care is required, active exercise is well tolerated, and full weight-bearing has been possible within three weeks. This device has furnished an effective means of arthroplasty or of reconstruction for non-union following fracture of the femoral neck.

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DISCUSSION

Dr. Robert Jaenichen, Saginaw, Michigan: A new type of metallic femoral-head prosthesis has been introduced. In 1944, the need was recognized for improvement in the treatment of certain types of pathological changes of the hip joint and, after careful thought and experimentation, the present prosthesis was devised. After study of the work of Dr. Austin Moore and Dr. Harold Bohlman, it was deemed practicable to develop this idea further. The head of the right femur of a dog was removed and a metallic prosthesis was substituted. Although the results were far from perfect, enough information was obtained to warrant further use of the device.

During the same period others were doing similar work, among them Dr. Bohlman, Dr. McBride, and Dr. Hudack. On a recent trip to Paris, I discussed this problem with Dr. Jean Judet, who has used acrylic prostheses in over 400 cases.

This discussion is based on forty-one cases with a follow-up period of more than one year, including seventeen males and twenty-four females. The ages ranged from thirteen to eighty-five years with an average of 51.7 years. Two cases were bilateral.

In this series of forty-three hips there were two cases of early and four cases of late postoperative infection. All infections have cleared. Two hips became subluxated; a shelf was created in one case of osteoarthritis, and the other case, that of a Charcot hip, stabilized itself by bone production about the prosthesis and acetabulum.

In four cases the head of the prosthesis became completely dislocated and required removal. Two of these cases were in the group of late postoperative infection. In the other two, the prosthesis was removed because it did not become stabilized in spite of prolonged immobilization in plaster. One of these, an old, unrecognized tuberculous hip with pathological dislocation, should not have been selected for this form of treatment. The other dislocation occurred in a case of rheumatoid arthritis. Arthrodesis of the hip has already been done or is planned in these four cases.

There have been no deaths in this entire series.

Mechanical failure of the prosthesis occurred in ten cases,—five in which the metal plate broke and five in which the screws broke.

No atrophy or absorption was noted about the prosthesis or screws. In fact, the reverse is seen. The bone becomes roentgenographically more dense in response to the stimulus of weight-bearing; this density is visible where the metallic head and neck bear against the bone, and also at the femoral cortex. In two cases with broken screws the plate became stabilized against the femur without reoperation, by deposit of dense bone beneath and around the plate, without any disability to the patient.

The reasons for these mechanical failures have been given by Dr. Peterson. Three different types of metal plates have been used. In eight cases the original type of plate was used; of these, three (37.5 per cent.) broke. An entirely new and stronger plate was then designed. It was tested in a walking machine by subjecting it to a stress equivalent to that exerted by a 250-pound individual jumping at the rate of thirty-six times per minute for over 1,000 consecutive hours, or two million steps. This type of plate has been in use for about one year. Not a single plate or screw has broken. This speaks well for the engineering skill of Mr. J. G. Collison.

Classification of Pathological Changes of the Hip Joint

	No. of Cases
1. Fractures of the femoral neck	
a. Ununited	11
b. Acute (primary replacement)	2
One of these patients was eighty-five years of age and a Smith-Petersen nail would probably have been inadequate because of the short subcapital fragment. She made a spectacular recovery and is still doing well.	
c. Aseptic necrosis	6
2. Old slipped epiphysis with deformity	1
This hip is stable and painless after failure following use of a Vitallium cup.	
3. Osteo-arthritis	7
Good results 3	
Poor results 4	
4. Rheumatoid arthritis	3
Good results 2	
Failure 1	
5. Marie-Strümpell arthritis	3
6. Old quiescent tuberculosis with pathological dislocation	1
7. Charcot hip	1
The operation is definitely not indicated in this type of case.	
8. Old healed septic hip and osteomyelitis	6
Good results 3	
Poor results 3	

It is difficult to evaluate the results in a group of patients with so many factors to be considered. These cases are distributed over a wide geographical area and represent many different diagnoses. In general, the result may be considered good if pain has been relieved, stability and motion have been restored, and the patient has been rehabilitated to a degree of usefulness and activity in keeping with his age and previous capabilities. If these goals have been only partially attained, the result is fair; if they have not been attained to any appreciable degree, the result is poor or a failure. On this basis there are twenty-three (56 per cent.) good results, eleven (27 per cent.) fair results, and seven (17 per cent.) failures. The cause of most of the failures was continued pain with instability or dislocation and, in two cases, infection requiring removal of the prosthesis. One patient with painful aseptic necrosis was walking very comfortably with a below-the-knee prosthesis and a femoral-head prosthesis before he became disabled because of a broken plate.

The number of cases in each pathological classification is small and, therefore, no final evaluation can be placed on each group separately. Fractures or non-union of the femoral neck unquestionably show the best results:

The reasons for good results are:

1. Careful selection of cases.

2. Proper and meticulous assembly of the prosthesis with the metal head resting on the stump of the femoral neck.

3. Restoration of the femoral-neck length to strengthen the gluteal leverage mechanism.

The reasons for poor results are:

1. Improper selection of cases, especially those fraught with the danger of dislocation or recurrent infection.

2. Errors in operative technique and in assembly of prosthesis.

This work is still in the developmental stage and much research remains to be done. Many important lessons have been learned and further improvement in design and technique probably will follow. The known mechanical defects of the prosthesis have been diligently corrected. The ultimate usefulness of this type of prosthesis will depend entirely upon its long-term successes.

In carefully selected cases we may reasonably expect:

1. Relief of pain in a large percentage of cases, especially in ununited fractures of the femoral neck.

2. Motion approaching normal in the presence of normal muscles and acetabular cartilage.

3. Stability, when normal length of the femoral neck has been obtained.

4. Compatibility. The present prosthesis is inert in the tissues and is well tolerated. In the few cases in which reoperation was necessary, the acetabulum has been found to be very smooth and shiny, without any corrosion of the metal or discoloration of the tissues.

5. Early ambulation and shortened hospitalization, even in the aged.

DR. LEO MAYER, NEW YORK, N. Y.: A little over two years ago, when I first heard of Dr. Jaenichen's work, it appealed to me at once as a very daring and courageous approach to a problem which, up to that time, had not been answered,—namely, what to do with ununited femoral-neck fractures in which there was extensive degeneration of the head. The only operation that I knew of was some type of osteotomy which would, as Dr. Smith-Petersen has pointed out, give distorted hip function.

This operation seemed to open up a possibility of restoring normal hip function; therefore, during the past two years, I have performed this operation in five cases. I am not entirely satisfied with the results in a single case, and yet I feel greatly encouraged by my observations in these cases.

The first patient was a man of seventy-two years with an ununited fracture of the femoral neck. He was walking well, four weeks after the operation. The Trendelenburg sign was negative. He had over 100 degrees of flexion of the hip and a normal range of abduction; he could abduct against gravity and had considerable agility. Unfortunately, the original type of plate was used in which there was a definite weak spot at the angle. About two months after the operation, he cracked his plate and a secondary operation was required.

In the last patient whom I operated on, the procedure was carried out four weeks ago in Israel. This man was one of two survivors of the Warsaw ghetto, where he had sustained a fracture of the femoral neck. This had remained ununited, and there was extensive degeneration of the femoral head. Within one week after the operation, this man was moving his hip in a normal fashion, and it took actual physical strength to keep him in bed for a two-week period. He is now walking on crutches—I don't permit full weight-bearing until a month has elapsed—but the chances are that he will have a practically normal hip.

I feel that Dr. Jaenichen has done a remarkable job and I am grateful to Mr. Collison also for the splendid work that he has done in developing this prosthesis into an apparatus which is mechanically sound.

DR. J. E. M. THOMSON, LINCOLN, NEBRASKA: Dr. Smith-Petersen, with years of experience, has proved the effectiveness of a metal prosthetic device in the hip. His Vitallium cup amounts to just that. The type of prosthesis offered by the speakers is slightly different from the Smith-Petersen cup and seems to be equally effective. Others have used similar adaptations. All involve a very extensive surgical procedure and destruction of a large portion of the upper end of the femur.

In 1948, I was impressed by a series of cases in which lucite prostheses had been placed obliquely through the trochanteric region. More recent reports give even more favorable results.

It seemed to me that, if the Smith-Petersen nail could hold the head of a fractured femur in place, it might not desecrate this useful intramedullary nail to embed it in a plastic head and let it hold the plastic head as an artificial prosthesis on the neck of the femur. To be useful for old people with non-union and complicated pathological changes in the hip, the method should be extremely simple. Usually much of the neck has been destroyed and only the base remains. When the prosthesis which I have developed is placed obliquely downward in a valgus relationship through what is left of the neck of the femur, with the Smith-Petersen nail element impinging well into the lateral cortex, one has solid fixation which is increased by weight-bearing. The stress and strain fall upon the lower portion of the neck of the femur, which is the strongest portion from an architectural standpoint. The end of the nail is embedded into the heaviest portion of the shaft on the lateral aspect.

Our series so far consists of fifteen patients between seventy and ninety-two years of age. The average time for performing the operation is about thirty to forty minutes. One death has occurred in the group, but this was one month after operation and was due to pneumonia. The first patient, aged eighty-five, was operated upon only five months ago, and she works in the garden every day. We have had one complication in the form of a dislocation, but if one will keep the leg in abduction and internal rotation for three weeks after operation, the possibility of dislocation is not great. We have found the posterior approach of Gibson or the lateral approach of Zahradníček to be the most advantageous method of applying this prosthesis.

These various methods seem to be deserving of further study.

DR. PAUL B. STEELE, PITTSBURGH, PENNSYLVANIA: In the last three months, we have treated eleven patients with a prosthesis which is similar except that the head element is screwed on, the screw being transfixed by small screws; the plate is thicker.

One thing that has not been stressed sufficiently is the severity of the operation; it is not a simple procedure. The Smith-Petersen cup operation is easy compared to this.

It is too early to evaluate our results.

DR. ALAN DEFOREST SMITH, NEW YORK, N. Y.: I would like to report upon the results in the cases that Dr. Hudack has treated in New York. His first operation, done nearly five years ago, was on a patient who had a low-grade chondrosarcoma of the upper end of the femur and otherwise would have been subjected to a hip disarticulation. We are restricting the operation to cases of this type because I feel that the operation is not physiological, and a long time must elapse before we can evaluate the results. It should not

(*Continued on page 102*)

LESIONS OF THE MUSCULOTENDINOUS CUFF OF THE SHOULDER

IV. Some Observations Based Upon the Results of Surgical Repair

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One hundred consecutive patients in whom shoulder-cuff repair was done were studied. These patients were followed for one to eleven years subsequent to repair, for an average exceeding three years. Coincident comparable studies of other common shoulder conditions are to be reported in detail elsewhere, but they will be drawn upon occasionally in the following discussion to illustrate and amplify certain points of interest. An attempt has been made to clarify some of the common discrepancies between certain established theories and postulates, and the behavior of the shoulder mechanism in clinical practice. Although the musculotendinous cuff was the primary subject of study, some information concerning the aberrations and functional significance of other components of the shoulder has also been obtained.

Concerning the Acromion

A serious effort to repair ruptured tendon cuffs was started in 1935. Originally exposure of the subacromial structures was obtained by modifications of Codman's saber-cut approach [4]. The divided acromion fragments were re-approximated and fastened together by sutures through drill holes in the bone. The results, in terms of postoperative pain and early availability of joint motion, were poor. Gradually, there evolved the practice of maintaining approximation of the acromial fragments by suture of the surrounding soft parts only. Apparently this technique allowed some mobility of the outer fragment of the divided acromion during the early postoperative period. No non-unions occurred, and the results improved appreciably and consistently.

Since 1938, the acromion has been divided transversely and the outer fragment has been excised and discarded in almost every case. The size of the discarded fragments was governed by the extent of subacromial exposure required, and they varied from a thin sliver of bone to the entire acromion. In muscular patients, it was difficult to palpate the resultant bony defects, regardless of the amounts removed. Except in thin patients, who had a large fragment removed and had no remodeling of the stump edges, no visible defects occurred. No subsequent functional defect was encountered in any of the cases. In several instances, division of the acromion inadvertently resulted in disarticulation at the acromioclavicular joint. Although disarticulation is considered neither advisable nor necessary, it should be reported that it was not found to have any harmful effects in these cases.

The late results commonly were characterized by small irregular outgrowths of bone from the stump of the acromion. Although present in the roentgenograms of most cases, these phenomena were not productive of symptoms in any case of transverse osteotomy. In one patient, the anterior, rather than the outer, half of the acromion was removed. The early result was good, but within a year bony outgrowths from the anterior edge of the stump created sufficient trouble to require a second, more extensive acromioplasty.

The acromion has not been divided for the removal of a calcific deposit from the rotator tendons. Several hundreds of such deposits have been removed simply and easily through a small, high, deltoid-splitting incision, rarely exceeding three centimeters in

length. The addition of dividing the acromion multiplies this operative insult to the tissues threefold, which would justify the expectation of a corresponding increase in the duration of disability and the difficulties attendant upon recovery. The results in the cases mentioned were uniformly excellent [10], and hospitalization seldom exceeded three days. Recovery of function was rapid, provided the coraco-acromial ligament was divided. The uniform excellence of these results substantiated the conviction that the removal of the acromion was unjustified, unnecessary, and ill-advised in operations for excision of a calcific deposit under ordinary circumstances.

The acromion was removed in a series of "stiff" shoulders. In the first few cases, this appeared to be a promising procedure. However, it was soon noted that preparation of the patient and the shoulder for operation entailed considerable passive mobilization of the extremity under anaesthesia. In subsequent cases, no improvement in glenohumeral motion followed excision of the acromion, provided care was taken to prevent manipulation of the arm prior to its removal. Following operative mobilization of stiffened shoulders, the results in patients who had the acromion removed were about the same as in those control cases in which the acromion was left intact.

The following conclusions seem permissible:

1. The acromion has little or no functional significance;

2. It may be divided or removed without fear of a functional defect, and with little or no risk of producing any significant cosmetic defect;

3. Excision of the acromion, *per se*, does not increase the available motion in a stiff shoulder;

4. Excision of the acromion is unnecessary and rarely justifiable for the removal of a calcific deposit.

Repair Following Removal of the Acromion

As emphasized by Smith-Petersen, excision of the acromion should be followed by a strong repair of the deltoid origin to the tissues overlying the acromial stump. The deep surface of the central portion of deltoid origin consists of a strap of tendon tissue usually three to five millimeters thick and two to five centimeters wide. It is essential for this tough tissue to be incorporated in the suture line. Sutures through deltoid-muscle substance alone were ineffectual, unless the shoulder was immobilized in abduction until complete healing occurred. When this technical prerequisite was overlooked, the usual result was a visible and palpable defect in the contour of the shoulder where the deltoid fibers had pulled away from the suture line. In three such cases this produced an obvious cosmetic blemish and some mild but permanent deltoid weakness.

The Coraco-Acromial Ligament

This structure was observed to be functionally unimportant in the normal shoulder and a major offending agent in most painful shoulders. In the great majority of patients in this study, and in many others operated upon for other conditions, this ligament was divided just lateral to the acromioclavicular joint. This procedure served a dual purpose, — to facilitate exposure of the subacromial region and to minimize the postoperative compression of the subacromial structures otherwise exerted by the sharp anterior edge of the ligament. No functional defects or instability of the glenohumeral articulation ensued. Postoperative pain was reduced markedly, and the recovery of shoulder motion was accelerated as a constant result, especially after operations for cuff repair and removal of calcific deposits. Consequently, division of the coraco-acromial ligament has become a routine step in all operations upon the subacromial structures, in which the acromion is left intact or only a small portion of its outer rim is removed.

For a long time it was not fully appreciated that the division of the acromion, with or without removal of the outer fragment, required a division of the coraco-acromial

ligament. The marked reduction in postoperative pain following a division of the coraco-acromial ligament without removal of the acromion, as compared to similar previous cases in which the ligament was not divided, was sufficient to permit the conjecture that the success of acromioplastic procedures [13] in relieving pain may in large part be dependent upon the release of tension resulting from division of this ligament.

The coraco-acromial ligament proved to be an essential factor in the etiology of many so-called "snapping" shoulders. The majority of such syndromes fell into two pathological categories. One resulted from a primary irritative focus producing bursal inflammation, followed by a heaping up of the thickened bursal roof into folds which snapped back and forth under the edge of the ligament on motion of the arm. The other consisted of some irregularity in the floor of the bursa,—for example, a quiescent deposit or a small and otherwise insignificant cuff tear, so situated as to snap under the edge of the ligament on appropriate motions of the arm. In the first group, snapping symptoms occasionally recurred following bursectomy alone. No recurrences followed bursectomy accompanied by a division of the ligament. In the second group, surgical eradication of the irregularity in the floor of the bursa relieved the snapping symptoms, but postoperative pain was usually severe and disability prolonged, except when the coraco-acromial ligament was also divided.

The Tendon of the Long Head of the Biceps

The majority of the ruptures of this tendon occurred at, or just proximal to, the upper extremity of the bicipital groove and were preceded by a cuff rupture which left this portion of the tendon exposed to trauma by the acromion on motions of the arm. Similarly exposed, unruptured but swollen, inflamed, and degenerating biceps tendons were commonly found to accompany cuff tears. A rupture of the long tendon of the biceps as an isolated lesion proved innocuous; while a rupture, accompanied by serious symptoms or protracted disability, proved to be a dependable indication of a significant cuff tear. Repair of the biceps in the latter circumstance proved neither necessary nor advantageous. Relief of symptoms and disability proved to be dependent upon adequate repair of the accompanying cuff rupture.

The mobility of the biceps tendon, in its groove, and in the shoulder joint, under various conditions of trauma and disease, and in shoulders approaching normal, was variable enough to preclude any description of the normal. Neither subluxation nor dislocation of this tendon from its groove was encountered, except when made possible by a tear of the transverse ligament or of the adjacent tendon at the cuff insertion. Although often suspected, both before and at operation, bicipital tenosynovitis, confirmed by microscopic study, was not encountered as an isolated lesion, but only as an extension of some inflammatory process originating in the shoulder joint.

The long biceps tendon was separated from its glenoid attachment and the intra-articular portion was withdrawn from the joint cavity many times. In stiff shoulders and in many cuff ruptures, the intra-articular portion was excised and the distal segment was anchored in the bicipital groove. In appropriate cuff tears, as previously described [11], the intra-articular portion was split; the two halves were used as an autogenous suture for cuff repair. Neither instability of the glenohumeral joint nor functional defects of the shoulder as a whole were observed to result from ablation of the intra-articular portion of this tendon.

The Glenoid Labrum

Lacerations, avulsions, and dislocations of this structure were observed in many shoulders with no history of dislocation. An intact labrum was not uncommonly found at an operation for recurrent dislocation. Several bucket-handle tears of this meniscus were encountered as isolated lesions. These produced internal derangements of the shoulder

mechanism, which were difficult or impossible to differentiate clinically from a "snapping" shoulder, a small cuff tear, or a recurrent subluxation of the humerus. The labrum was partially excised many times, especially in operations for the repair of a recurrent dislocation. No functional defects ensued. It appeared reasonably certain that the glenoid labrum, *per se*, was an insignificant factor in recurrent dislocation [1] and had little functional significance in its own right.

The Subdeltoid Bursa

The diameter of this bursa varied from two and five-tenths to ten centimeters, with an average of about five centimeters. Stringlike, incomplete loculations, appearing at first glance to be adhesions, were found with equal frequency in both normal and diseased bursae. We concluded that they were nothing more than developmental anomalies. Occasionally the loculations were complete; so that two or more separate bursal cavities coexisted. No pathological adhesions were encountered except as the result of trauma or of infection in the bursa or as the result of chronic inflammation centered around a calcific deposit. However, it has been repeatedly demonstrated that the immature or careless surgeon can be misled by false dissection into making an incorrect diagnosis of "adhesive bursitis". This is especially true in the exploration of stiff shoulders in which the bursal walls are thin, anaemic, and atrophied.

In the presence of full-thickness cuff tears, the bursal walls were consistently thickened and somewhat inflamed. In long-standing lesions, they were leathery and avascular, due to fibrous-tissue replacement. Because of a spreading apart of the rent in its floor, the bursal diameter and capacity were also increased in the presence of a major cuff tear. Joint fluid entering through the rupture increased the quantity of fluid in the bursa, which, under normal circumstances, was just sufficient to supply lubrication. Any appreciable amount of fluid encountered upon opening the bursa, in the absence of acute inflammation, proved pathognomonic of cuff rupture, whether or not such a lesion was immediately apparent. If, with the bursa open in the presence of a cuff tear, the capacity of the shoulder-joint capsule was decreased by abduction, a gush of joint fluid into the bursa was the rule. This phenomenon was only occasionally demonstrable by clinical examination prior to operation, but when present proved a pathognomonic sign of rupture. As a result of its increased size and fluid content, the bursa appeared to fulfill its lubricating function more efficiently than normally when a full-thickness cuff rupture was present. Occasionally, the synovial floor of the bursa remained intact, covering a rupture through the full thickness of the underlying tendon fibers. In such cases, the tendon defect was always palpable, even if not immediately visible.

In stiff shoulders, the bursa consistently appeared smaller and drier than the normal. Its walls were usually thin, anaemic, and atrophic. In such cases, false dissection, suggestive of adhesive or obliterative bursitis, was a common fault. However, when the true bursal cavity was examined, its appearance was usually compatible with disuse rather than intrinsic pathology. Neither inflammation nor adhesions were encountered in the bursae of stiff shoulders as a general rule. The two exceptions to this statement were in those lesions primarily caused by trauma, infection, or prolonged inflammation at the site of a calcific deposit. In other stiff shoulders, operative observations permitted the conclusion that these abnormalities in the subdeltoid bursa were the result, rather than a cause, of stiffness.

No anomalous communications between the subdeltoid bursa and the shoulder joint were encountered. All communications appeared to be the result of degeneration or trauma, or both. In 1938, in conjunction with Dr. Paul C. Swenson, arthrography was utilized to substantiate the clinical diagnosis of rupture. Thirty to fifty cubic centimeters of filtered oxygen were injected into the shoulder-joint cavity under fluoroscopic control. In the presence of a full-thickness tear, it was possible for the observers to watch the gas escape

through the tendon defect into the bursal cavity. All cuff ruptures indicated by arthrography were confirmed by operation. Subsequently, this procedure became unnecessary to diagnosis and was discontinued for reasons to be discussed later under diagnosis and indications for repair.

The Musculotendinous Cuff

This structure which contains the conjoined tendons of the subscapularis, supraspinatus and infraspinatus, and teres minor, and which fuses with the lateral extremities of the glenohumeral and coracohumeral ligaments, has been the subject of some argument and confusion with respect to its functional relationship to the deltoid muscle [6]. The following constant findings helped to clarify the true role of the cuff in elevating the arm:

1. No patient who had a complete or massive avulsion of the cuff from the humerus was able to initiate abduction of the arm against gravity in any position. When the arm was passively placed in abduction, none could hold it there in any position. Contraction of the deltoid served but to pull the humeral head closer to the acromion. It was beyond doubt that some function of the cuff unit was necessary to the elevation of the arm.

2. Those patients who had a complete tear of the supraspinatus tendon alone were able to initiate and maintain elevation of the arm against gravity in all positions. It was obvious that, contrary to popular belief and despite the continued postulates of many standard texts, the supraspinatus function, *per se*, was not necessary to the initiation or maintenance of abduction.

3. No patient with a rupture less than three centimeters in extent, regardless of the location of the tear or the tendons involved, lost the ability to elevate the arm. Operative findings, correlated to preoperative examinations, indicated that the function lost from a rupture less than three centimeters wide was compensated for by the adjacent cuff attachments in much the same way as intact vastus expansions may compensate for the loss of knee-extension power resulting from a fracture of the patella.

4. All patients with a tear wider than three centimeters, regardless of location, and in a degree varying with the size of the tear, were unable, or barely able, to maintain the arm elevated against gravity or the resistance of one finger; but this was true only when the arm was rotated into a position where the maintenance of elevation created a functional demand upon the torn portion of cuff. The correlation of operative and preoperative findings demonstrated that elevation required the function of that segment of the cuff superior to the humeral head and parallel to the plane of elevation. The supraspinatus is widely accepted to be this essential force. It is an anatomical fact, however, that any portion of the cuff from the subscapularis to the teres minor may become the intrinsic motor force essential to elevation, depending upon the rotational position of the humerus and its plane of elevation. This was confirmed repeatedly in cases with ruptures exceeding three centimeters by demonstrating that the patient could not maintain elevation against

TABLE I

DIAGNOSIS

Operative Repair (100 Cases)	Massive Avulsions (18 Cases)	Complete Tears (66 Cases)	Incomplete Tears (16 Cases)
Average age	57 (41 to 74)	51 (19 to 72)	41 (16 to 63)
Male	16	42	9
Female	2	24	7
Heavy work	12	28	5
Light work	6	37	11

TABLE II

THE SYMPTOMATIC RESULTS

Operative Repair (100 Cases)	Massive Avulsions (18 Cases)	Compete Tears (66 Cases)	Incomplete Tears (16 Cases)
No symptoms of any kind	10	38	8
Occasional ache in bad weather the only complaint	5	23	7
	15	61	15
Some complaint of pain	3	5	1

gravity when the rupture was superior to the humeral head, but could do so with ease when the arm was rotated so that the rupture assumed a position behind or in front of the humeral head. Preoperative clinical evaluation along these lines has made it possible to demonstrate not only the presence of, but also the approximate extent of, the tear and the tendons involved in the majority of cases.

Some information was obtained concerning the role played by the intrinsic muscles in suspending the humerus. Anatomically it may be deduced that the humerus is suspended from the scapula by the cuff, and deltoid, triceps, and coracoid muscles. Of the cases studied, eighteen had massive or complete avulsions of the cuff from the humerus. About 50 per cent. of these were in elderly patients with weak or debilitated musculature. Inferior subluxation of the humerus was present in the latter cases, despite intact extrinsic muscles. The majority applied for help, complaining of neurological disorders in the hand or forearm, resulting from undue traction upon the brachial plexus. Cuff repair not only relieved the neurological disturbances, but also the humeral subluxation. In similar younger patients with strong extrinsic muscles, inferior subluxation of the humerus was not encountered. It appeared that the cuff contributed significantly to suspension of the humerus, but that its loss could be well compensated for by extrinsic muscles of good quality.

Concerning the Diagnosis of Cuff Tear

The literature contains much about the diagnosis of cuff rupture which has proved at variance with clinical observations. Apparently, certain widespread concepts were based on theories originating in the dissecting room or from postmortem observations. The living shoulder proved to be a complicated, but at the same time a labile, mechanism, well able to compensate for many gross physical defects. The discrepancies of some of the widely accepted diagnostic concepts were well illustrated by this study:

1. It has been accepted as almost axiomatic that cuff rupture is a condition peculiar to males who do manual labor and who are past forty years of age. The fallacy of this as a diagnostic criterion is shown in Table I. However, it must be stated that the ease of rupture appeared to vary directly with the age and the amount of wear and tear of the tendon tissue involved.

2. The tensile strength of normal cuff tendons is great, and consequently it has been widely misconceived that rupture requires a forceful trauma. As pointed out by Codman, this is not true. In this series, the majority of patients related some history of injury, but often the trauma was mild enough to escape notice or to be discarded as inadequate. The explanation of this apparent paradox became clear when it was found that almost every

rupture took place through degenerated tendon tissue and retracted by a splitting apart of the more normal proximal tendon fibers [7]. A few patients could recall no trauma of any kind and had ruptures that appeared to be entirely the result of degenerative changes. No transverse rupture through normal healthy tendon was encountered, nor was any healthy tendon avulsed from the bone, except by severe trauma.

3. Many surgeons exclude the possibility of rupture when the history reveals shoulder symptoms prior to the injury in question. The fallibility of this practice was demonstrated, since many times operation disclosed evidence of a recent extension of a long-standing rupture, in which there was shown that the lesion had been compensated for until the tear was enlarged by the last injury.

4. Most surgeons do not consider the possibility of cuff rupture seriously, unless the patient has great difficulty or is unable to elevate the arm. This sign is present to some degree in most shoulders painful from any cause. As previously stated, many patients with small cuff ruptures had little or no difficulty in elevating the arm.

5. The diagnosis of rupture is commonly made in patients who have a stiff shoulder. No patient in this series had a stiff shoulder; no stiff shoulder explored to date had a torn cuff.

6. It is widely accepted that the diagnosis of a cuff rupture can be made only if no changes are evident in the roentgenogram. In some instances to be mentioned, the only reliable evidence of rupture was in the roentgenogram. In the great majority of cases, confirmatory evidence of pathological changes at the point of rupture was shown by roentgenogram in the form of degenerative changes in the bone. This finding supported the diagnosis consistently enough to warrant notice as a positive clue in doubtful cases.

Many other signs and symptoms generally credited as evidence of a cuff tear were found to be similarly misleading or fallacious. Certain criteria [9], outlined below, proved constant, dependable, or pathognomonic. Only two constant signs of value to the differential diagnosis were established. These were present in every case:

1. The absence of restriction of passive motion. The shoulder containing a torn cuff often seemed stiff when moved against gravity. This was usually due to pain. However, the presence of unrestricted glenohumeral motion was demonstrable without undue discomfort by asking the patient to bend over slowly and try to touch his toes.

2. Selective myopathies. Within two weeks of rupture, atrophy was visible and atony palpable in the spinati bellies. The deltoid, if abnormal at all, usually was hypertrophied in contradistinction to the deltoid atrophy common to most other conditions of painful shoulder.

Four dependable criteria were established. These were usually demonstrable:

1. The absence of a calcific deposit in the roentgenogram. Several hundreds of such deposits have been excised. Only twice did a cuff rupture and a calcific deposit co-exist. In each case the rupture was a small, degenerative lesion, unassociated with the deposit, and of no clinical significance. The presence of a deposit, therefore, is concluded to be

TABLE III

THE FUNCTIONAL RESULTS

Operative Repair (100 Cases)	Massive Avulsions (18 Cases)	Complete Tears (66 Cases)	Incomplete Tears (16 Cases)
No motion defect	4	44	8
Elevation defect of less than 20 degrees	7	17	6
Elevation defect of more than 20 degrees	7	5	1

TABLE IV

DISABILITY TIME

Operative Repair (100 Cases)	Massive Avulsions (18 Cases)	Complete Tears (66 Cases)	Incomplete Tears (16 Cases)
Average preoperative disability	8 weeks	12 weeks	16 weeks
Average postoperative hospital days	17	14	15
Average return to light work	13 weeks	8 weeks	8 weeks
Average return to regular work	16 weeks	13 weeks	10 weeks

strong evidence against a tear. In utilizing this as a negative diagnostic point, it is important not to misinterpret an avulsed fragment of a tuberosity, or calcification, in a retracted tendon stump as a collection of amorphous calcific material.

2. Palpation of the rupture. A complete-thickness rupture was almost always palpable as a hiatus in the tendon cuff. The ability to demonstrate this sign required considerable practice and, above all, a completely relaxed deltoid; but, once it was established, it proved to be the most dependable criterion available.

3. Crepitus. The presence of a characteristic soft crepitus, occurring when the retracted tendon stump was rolled between the palpating finger and the humeral head, proved to be a frequent dependable sign.

4. Painless, or only mildly uncomfortable, inability or definite weakness in maintaining abduction of ninety degrees against gravity or light resistance, in the presence of intact axillary and suprascapular nerves. The physiological basis of this finding has already been discussed in relation to cuff function.

Only two pathognomonic criteria were encountered:

1. Arthrography, which has been mentioned previously.

2. Displacement of a fractured tuberosity fragment. When either tuberosity was fractured, it was found to remain in apposition to the humeral head only by virtue of the continuity of cuff fibers inserting into both the fragment and the parent bone. Gross displacement invariably signified gross cuff rupture. Persistent significant displacement (one centimeter or more) proved to be an indication for operative repair at the earliest possible moment.

INDICATIONS FOR TREATMENT

At the beginning of this study, it was considered that early repair was indicated in all cuff ruptures; however, evidence was accumulated to indicate that in many cases no repair was needed, and that in only a few was immediate repair required. Some of this evidence is as follows:

TABLE V

ECONOMIC REHABILITATION

Returned to Work	Massive Avulsions (18 Cases)	Complete Tears (66 Cases)	Incomplete Tears (16 Cases)
Regular work	12	59	12
Light work only	3	4	1
Record uncertain	3	3	3

1. Twenty to 30 per cent. of unselected cadaver specimens (past sixty years of age at death) were found to have had cuff tears [12]. Obviously, one-quarter of all persons past sixty do not need operation, so that the remaining intact shoulder mechanism must compensate for the cuff injury in many instances.

2. The preceding postulate was confirmed by the fact that, after the enthusiasm for early repair had given way to a more mature consideration of its practical necessity in each case, many patients were found to recover spontaneously if given the chance. Most of these patients retained the signs, but gradually lost all, or almost all, the symptoms and the disability produced by the lesion.

3. Almost all ruptures occurred through degenerated tendon tissue. The torn or avulsed margins were fibrotic and avascular. In short, the moment it occurred, the rupture was the equivalent of a chronic, long-standing lesion. Anatomical reposition of the fragments and hope for healing by first intention were precluded from the start, and, consequently, there was no particular advantage to be gained from early repair.

4. During the early period following injury, pain makes an accurate anatomical diagnosis difficult; and the risk of doing an unnecessary exploration is increased appreciably.

5. The results of early and late repair were identical, except for delays in recovery resulting from weakness and debility due to disuse of the uninjured portions of the shoulder mechanism in certain long-standing cases.

These and other practical points resulted in a strong conviction that early repair is contra-indicated in all except those cases with massive damage, in which there is early evidence that severe symptoms and disability will persist. Experience indicated the wisdom of a minimum period of two weeks' observation [3] after which, if symptoms are unabated and the diagnosis remains definite, repair should be considered. If, after two weeks, improvement is noted, conservative therapy is continued until the symptoms and disability reach a stationary level. At that time repair is contemplated, not because a cuff rupture has been demonstrated, but upon a basis of the persistent symptoms and disability.

Certain small cuff tears could not be diagnosed with certainty. In these cases a presumptive preoperative diagnosis was made by the exclusion of other lesions compatible with the clinical picture. More than a few cuff tears were found at operation in the absence of any preoperative diagnosis. The number of such experiences in connection with disabled shoulders in which no preoperative diagnosis other than "internal derangement" was possible demonstrated exploration to be amply justified in selected cases. Adequate selection required: (1) the exclusion of a systemic disease which involved the shoulder only as a local manifestation, (2) a reasonable certainty that the derangement was of mechanical origin and situated in the subacromial mechanism, and (3) symptoms or disability sufficiently severe or prolonged to justify surgical intervention from the patient's standpoint. The results were very satisfactory. It is now concluded that the preceding three prerequisites constitute an indication for exploration of the subacromial mechanism with or without a definite anatomical diagnosis.

TREATMENT

At the beginning of the study, it was believed that the position of abduction facilitated the healing of a cuff tear. However, the accumulation of evidence demonstrated this so-called "conservative abduction treatment" to be not only unnecessary, but also harmful to the healing of the rupture, to the shoulder mechanism, and to the patient as a whole. Examination of ruptures at operation disclosed that:

1. When the tear was through the incomplete thickness of the cuff, the remaining intact tendon fibers held the ruptured fibers in place, regardless of the position of the arm;

2. Abduction rarely approximated the separated margins of a complete-thickness rupture;

3. Abduction usually compressed the retracted tendon stump between humerus and acromion to the further detriment of its already impoverished blood supply;

4. Abduction constantly pumped joint fluid through the tear into the bursal cavity. The anticoagulant properties of such a synovial-fluid bath are a deterrent to connective-tissue healing;

5. Finally, previous experiences with treatments requiring prolonged immobilization of the abducted arm demonstrated that these treatments often had a penalty more severe than that of the untreated original lesion.

A program designed to control pain and maintain the mobility of the shoulder proved to be the most efficient form of conservative treatment. A sling and, if needed, a swathe were used during the acutely painful stages following injury. Both were discarded as soon as symptoms permitted. Appropriate palliative measures were employed as needed. Simple home measures of a palliative nature proved more efficient than formal physical therapy. Gradually, progressive active exercises and the resumption of use at frequent intervals up to, but not past, the limits of pain or fatigue, were insisted upon. Many patients in whom the diagnosis of cuff rupture was well established recovered on this regimen without further treatment. Those who failed to do so and were operated upon form the main basis for this report.

Operative Treatment

The surgical techniques for cuff repair have been described in detail elsewhere [5, 7, 8]. At the beginning of this study it was considered that repair should restore and maintain normal anatomy by replacement and fixation of the torn end of the tendon cuff to the point from which it had been ruptured or avulsed [14]. Attempts to attain this objective were usually predestined to failure. Operative experiences soon demonstrated that no two ruptures were identical or amenable to identical repair techniques, that each required a plastic tenorrhaphy suited to its own individual characteristics, that restoration of normal anatomy was relatively unimportant, and that there were only three essential prerequisites to a successful repair:

1. The snug apposition of healthy to healthy tissue, without tension, at the site of repair;

2. The restoration of continuity between the short rotator muscle bellies and the humerus;

3. The restoration of a smooth surface for articulation with the acromion (or removal of the acromion).

Postoperative Treatment

Only one of the 100 patients included in this study was immobilized following operation. It now seems probable that complete rest of the shoulder during the first week or two after operation might have been of some benefit. It was demonstrated beyond question that the prolonged postoperative immobilization employed prior to this study often did more harm than good. However, these cases, with the one exception mentioned, were managed by the following general postoperative program:

Rest and comfort were provided by sling and swathe, or balanced suspension, during the painful wound-healing period. Gentle pendulum exercises were started as soon as symptoms permitted, usually between the third and seventh postoperative days. The scope and frequency of active exercising were steadily increased, and a resumption of use encouraged up to the limits of pain or fatigue. The rapidity with which active motion and use of the arm against gravity were allowed varied according to the magnitude of the tear, the quality of the repaired tissues, and the mechanical efficiency and freedom from tension of the repair itself.

THE RESULTS

These patients were operated upon for disability resulting from pain, weakness or difficulty in function of the extremity, and inability to do their regular work. The fact that a large proportion were compensation cases is worthy of consideration in scrutinizing the results.

All patients retained an unrestricted range of passive motion. The mobility as shown in Table III refers to active motion against gravity. Defects in elevating the arm appeared to result from, and vary directly as, the alteration of the point of insertion of the cuff into the humerus,—for example, retracted large tears were reinserted into the humeral head at whatever point they would reach without tension, with the arm at the side.

Preoperative disability periods varied from a few days to a few years and, as previously stated, had no important bearing on the quality of the final result. Hospitalization was continued in each case, not only until the operative wound was well healed, but also until the patient was well enough established in his rehabilitation program to make continued improvement at home a reasonable expectation, even though it was not closely supervised. Relatively normal light use of the extremity was present in the majority of cases at the time of discharge from the hospital. The time of return to work for ordinary patients averaged only about 50 per cent. of the period necessary for compensation cases.

SUMMARY

Some observations of clinical interest and importance concerning the shoulder mechanism are reported, along with certain conclusions resulting from clinical experiences in the exploration and repair of injuries to the musculotendinous cuff. Appropriate references and credits to the work of Codman were so numerous as to make their individual documentation impractical. Suffice it to say that many of the major points which have been discussed serve but to confirm the previously published [4] postulates of this great student of the shoulder mechanism.

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SHELF OPERATION FOR CONGENITAL SUBLUXATION AND DISLOCATION OF THE HIP

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The term "congenital dysplasia of the hip" is used to describe a group of developmental malformations of this joint. The chief abnormalities consist of a shallow acetabulum, deficient in its outward prolongation, and of associated hypoplasia of the head of the femur. Hypoplasia and anteversion of the femoral neck and torsion of the shaft of the femur also have been noted in this condition. When the acetabulum is not sufficiently deep to accommodate the head of the femur in the weight-bearing position, there may be incomplete displacement (subluxation) or complete displacement (dislocation) of the head of the femur from the malformed acetabulum.

A detailed history of the treatment of congenital subluxation and dislocation of the hip is beyond the scope of this report. Advance in the knowledge of this disorder has been made through the efforts of physicians [3] working in diverse fields in many parts of the world. Although many methods of treatment have been devised, all the therapeutic problems of congenital dislocation of the hip have not been solved. Each of these methods has its indications for use which depend upon the age of the patient and the severity of the anatomical defect.

Manipulative reduction, if performed before the age of three years, may produce satisfactory function of the hips. Open reduction is more frequently required beyond that age. Operative correction of the torsional deformity of the neck and shaft of the femur becomes more important as the child's age increases. When the head of the femur is hopelessly deformed, trochanteric cup arthroplasty or high osteotomy of the femur may produce a functional hip. When the dislocation is irreducible or the deformity of the acetabulum or femur is so severe that the head of the femur will not be retained in its proper relation to the acetabulum, the shelf operation offers the possibility of increasing the functional stability of the hip. However, when this operation is performed on patients who are less than seven years of age, a high proportion of the newly constructed shelves become absorbed and disappear.

With each year that the deformity progresses untreated, the problem becomes increasingly difficult. The cases reported in this paper represent the results of only one form of treatment of a late phase of congenital malformation of the hips.

ETIOLOGY OF CONGENITAL SUBLUXATION AND DISLOCATION OF THE HIP

All the mesodermal structures of the hip may exhibit dysplastic features, and this is frequently the case. The hereditary background of this disorder was clearly shown by Faber's roentgenographic studies of the families of ten children with classic dislocation. Many authors accept the theory that the primary abnormality in congenital dysplasia of the hip is an intrinsic germinal failure of development of a portion of the acetabulum. Badgley concluded from his embryological studies that environmental factors, extrinsic to the hip and acting at the time of the rotation of the limb buds, may interfere with the adaptational remodeling of the acetabulum and head of the femur. He pointed out that, in complete dislocations, the head of the femur, lacking the stimulus for overgrowth seen in subluxations, remains small and round, although becoming flattened on the side if in contact with the ilium (Figs. 1-A and 1-B). Hart accepted both the genetic and biomechan-

FIG. 1-A FIG. 1-B

Fig. 1-A: Group 4*a*, congenital dislocation of the left hip, showing deformed, flattened head.
Fig. 1-B: Two years and seven months after operation, a good functional result was obtained.

ical theories and stated that a combination of these two forces may produce the primary acetabular malformation. Retracing the ontogeny of man, Böhm noted that the acetabulum of the human foetus has the same oval shape as the acetabulum of the gorilla; in both

FIG. 2-A FIG. 2-B

Fig. 2-A: Group 3*a*, congenital subluxation of the left hip before operation.
Fig. 2-B: Adequate shelf six weeks after operation. Eight years and eight months later the result was good.

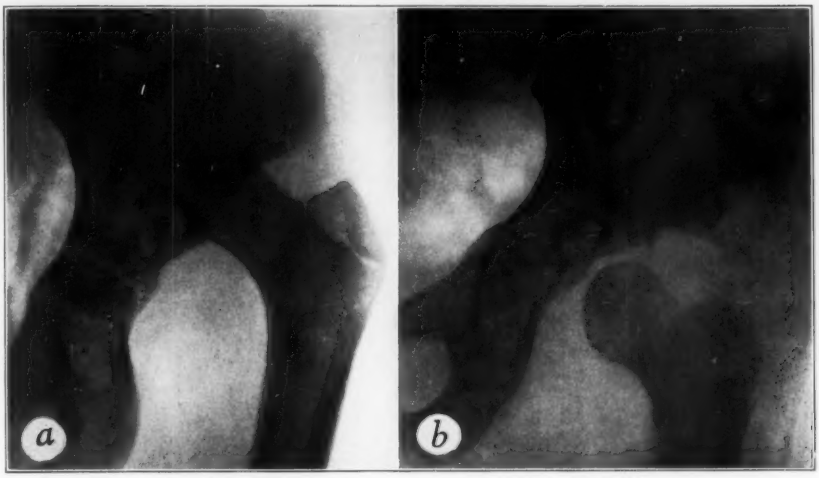

FIG. 3-A FIG. 3-B

Fig. 3-A: Group 4*b*, congenital dislocation of the left hip before traction and shelf operation.
Fig. 3-B: Two months after traction and shelf operation. Observation twelve years after operation disclosed that a good result had been obtained.

the posterior portion of the roof is lacking. He felt that this defect in the posterior superior buttress of the ilium is essential to the pathogenesis of congenital dislocation of the hip.

TECHNIQUE OF PERFORMING THE SHELF OPERATION

The purpose of the shelf operation is to afford a stable weight-bearing ledge to support the pelvis on the head of the femur (Figs. 2-A and 2-B).

In 1931, Ghormley described his method of performing the shelf operation. This procedure will be described in detail because it was used, with minor exceptions, in all the shelf operations reported in this study. Ghormley stressed the importance of (1) heavy traction for several days before operation or at the time of operation to reduce the dislocation or to bring the head of the femur as far down as possible (Figs. 3-A and 3-B); and (2) building a shelf which is large enough to cover the head of the femur without interfering with abduction by abutting against the trochanter.

The anterior portion of the iliac crest and the margin of the acetabulum are exposed through a lateral incision which is curved posteriorly.

With the head pulled down as far as possible on a traction table, chips of bone are turned down over the head, its capsule being kept intact. These chips are cut with a slightly curved chisel in such a way as to have their heaviest portion or base as close to the head or new acetabular margin as possible, and enough are laid down to have a fairly straight cut above the acetabulum. The anterior superior spine and the anterior portion of the iliac crest are freed entirely of periosteum and muscle attachments. This portion of the ilium is then cut away, with the chisel so held that the cut surface is beveled sharply downward (Fig. 6). The cut edge of this graft is then placed in the groove about the acetabulum and is driven inward, the beveled edge fitting snugly so that the entire shelf structure is held firmly in place (Fig. 6). The wound is closed. Traction straps having previously been fitted to the leg, a cast is applied to the ankle, and traction is maintained. After eight weeks, the traction is omitted and the patient is allowed to be up. Skeletal traction may be used if it seems indicated, in which case the cast should extend only to the knee. Walking with crutches may be commenced at the end of eight weeks, and without crutches at the end of twelve weeks, if the shelf seems sufficiently strong.

FIG. 4-A FIG. 4-B

Fig. 4-A: Group 3a, congenitally insufficient acetabulum before operation.
Fig. 4-B: Four years and two months after operation, including screw fixation of the shelf. A fair result was obtained.

FIG. 5

Fig. 5: Normal hip with a well formed acetabulum, head, and neck. The CE angle measured +37 degrees. It was formed by a perpendicular line through the center of the head of the femur and a line joining the outermost edge of the acetabulum with the center of the head.

Occasionally, to be absolutely sure that the shelf is at the desired level, the capsule is opened. Through a small incision, the margin of the acetabulum is inspected; the trough for the iliac graft is then cut at the optimal site. Most often the shelf can be quite firmly embedded; however, if it is not completely stable, one or two long Vitallium screws can be used to secure it to the pelvis (Figs. 4-A and 4-B). The desired curve to the shelf is usually present when the anterior iliac crest is used; but more curve can be obtained, if desired, by making transverse saw cuts through half the thickness of the ilium and green-stick fractures in the graft to close these saw cuts.

MATERIAL AND METHOD OF STUDY

The cases selected for this study included all patients (seventy-six) upon whom shelf operations had been performed because of congenital dislocation or subluxation of the hip during a ten-year period (January 1936 through December 1945) at the Mayo Clinic. Sixty-six of the operations were unilateral and ten were bilateral. Thus, a total of eighty-six shelf operations were reviewed. Nearly all of the frank dislocations were irreducible because of the age of the patient or because of previous unsuccessful manipulative or operative attempts at reduction.

Roentgenograms taken before operation were studied to classify abnormalities of the acetabulum and of the head and neck of the femur. Pronounced degrees of dysplasia and subluxation of the hip are easily recognized, but slight changes in the development of the roof of the acetabulum and its relationship to the position of the head of the femur are difficult to classify. The structural integrity of the hip for stability in weight-bearing depends upon these factors. In 1939, Wiberg reported a simple objective method for establishing this relationship, called the CE angle (*center* of femoral capital epiphysis to the *edge* of the roof of the acetabulum). The size of this angle is indicative of the development of the roof of the acetabulum as well as of the position of the femoral head in relation to its acetabular covering. Measurement of this angle was found to be useful in classifying various degrees of dysplasia and subluxation of the hips.

The CE angle is measured by drawing a line from the center of the head of the femur to the lateral edge of the roof of the acetabulum. The angle which this line forms with a vertical line through the center of the head of the femur is the CE angle. The vertical line is drawn perpendicular to a horizontal line across the pelvis, through the centers of the primary acetabula. The center of the head of the femur is the center of the arc described by the curvature of the head of the femur.

This procedure is simplified by using a transparent circular protractor, on the face of which are drawn several concentric circles (Fig. 5). The center of the head of the femur is the center of that circle which best encloses

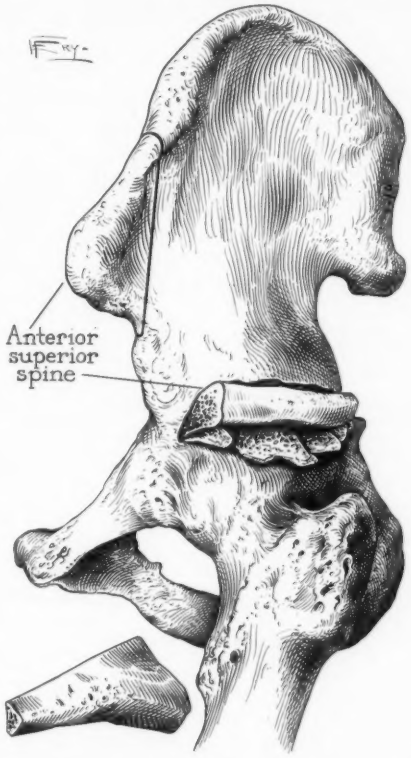

FIG. 6

Line of cut through the anterosuperior portion of the iliac crest. The cut edge is beveled and driven into the groove fashioned above the acetabulum[5].

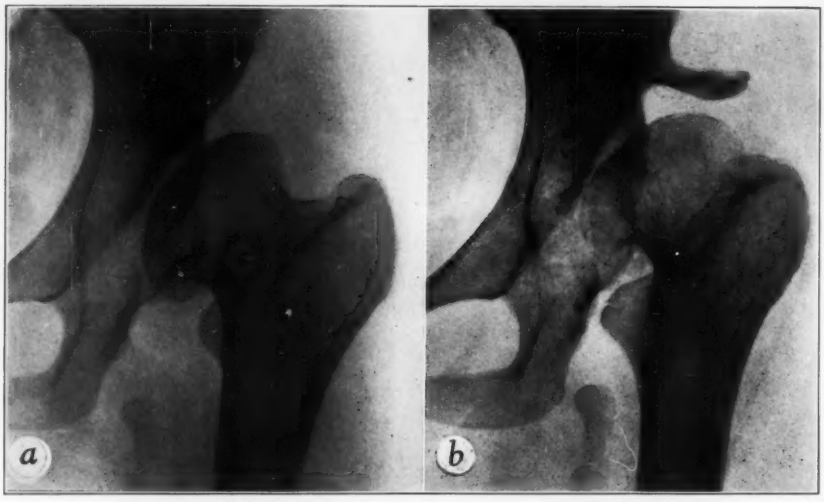

FIG. 7-A FIG. 7-B

Fig. 7-A: Group 3*b*, left hip, before operation.
Fig. 7-B: Two months after operation. A poorly shaped shelf, which was placed too high, is evident. The result was poor.

the head. The CE angle is read in either plus or minus degrees. When the head of the femur is not adequately covered by the roof of the acetabulum, the CE angle will be low or minus. The angle becomes less when the size of the acetabular roof decreases and when the head of the femur is displaced laterally. Thus, the CE angle is an expression of both the development of the acetabulum and of the position of the femoral head in relation to the acetabulum. This angle can be used for the diagnosis of both acetabular dysplasia and subluxation of the femoral head.

Severin measured this angle on the hips of 136 children between the ages of six and thirteen years. In this age group, he considered a CE angle of less than 15 degrees as an indication of a definitely pathological condition, values between 15 and 19 degrees as uncertain, and values of more than 20 degrees as normal. Severin accepted Wiberg's values for the CE angle for individuals over fourteen years of age. In this older age group, angles less than 20 degrees denote a pathological condition, angles between 20 and 25 degrees are not diagnostic, and angles of more than 25 degrees are normal.

Severin used these values to classify the abnormalities observed in the roentgenograms of 330 persons with congenital dislocation of one or both hips. The authors have modified

TABLE I

ANATOMICAL CLASSIFICATION ON THE BASIS OF ROENTGENOGRAPHIC FEATURES IN SEVENTY CASES (140 HIPS)

Classification	*a*	*b*
Group 1 (Normal)	34	3
Group 2 (Dysplasia)	8	5
Group 3 (Subluxation)	17	21
Group 4 (Dislocation)	24	28
Total	83	57

this classification and have divided their cases into the following groups, based on the anatomical features observed in the roentgenograms:

Group 1

Normal hips: well formed acetabulum, femoral head, and neck.

a. Normal. CE angle of more than 19 degrees for ages six to thirteen years, and of more than 25 degrees for age fourteen or over (Fig. 5).

b. Uncertain range. CE angle between 15 and 19 degrees for ages six to thirteen, and between 20 and 25 degrees for age fourteen or over.

Group 2

Dysplasia: deformity of femoral head, neck, or acetabulum, but no subluxation.

a. Mild. CE angle of more than 15 degrees for ages six to thirteen, and of more than 20 degrees for age fourteen or over.

b. Moderate. CE angle of less than 15 degrees for ages six to thirteen, and of less than 20 degrees for age fourteen or over.

Group 3

Subluxation: head is displaced upward and laterally and there is a break in Shenton's line.

a. Moderate. CE angle is slightly positive or equal to zero (Figs. 2-A, 2-B, 4-A, and 4-B).

b. Severe. CE angle negative (Figs. 7-A, 7-B, 8-A, 8-B, and 8-C).

Group 4

Dislocation.

a. The head articulating with a secondary acetabulum separate from the primary acetabulum (Figs. 1-A, 1-B, 9-A, and 9-B).

b. No secondary acetabulum present (Figs. 3-A and 3-B).

CLASSIFICATION OF CASES STUDIED

Of the seventy-six patients having shelf operations for congenital dislocation and subluxation of the hip, follow-up records were incomplete for six who had unilateral operations; these patients were excluded from the present study. Of the remaining seventy patients, fifty-nine were females and eleven were males. Their ages ranged from three to forty years. The average age at which the shelf operation was performed was eighteen years. No racial predominance was present. A history of congenital malformation of the hip in some other member of the family was given by five patients. In most instances, the disability was first noted when the child began to walk. Fifty-two patients (74 per cent.) gave a history of previous unsuccessful attempts at manipulative reduction (forty cases) or open operative reduction (twelve cases).

The roentgenographic features of both hips of each of the seventy patients were studied. Each hip was classified according to the method described previously; the results are indicated in Table I.

In the sixty cases in which a unilateral shelf operation had been performed, abnormalities of the opposite hip (not operated upon) were noted in twenty-three cases, or 38 per cent. Only thirty-seven (26 per cent.) of the entire group of 140 hips studied (seventy cases) could be classified as completely normal.

In this study, the functional status of each hip was estimated from the records of examinations made before and after the shelf operation had been performed. No attempt was made to evaluate the result of the operation in a case in which the follow-up period was less than one year. The follow-up period for the seventy patients varied from thirteen months to thirteen years and two months; the average duration was four years and eight

TABLE II

PREOPERATIVE ANATOMICAL CLASSIFICATION OF SIXTY HIPS IN WHICH UNILATERAL OPERATION WAS PERFORMED

Classification	No. of Cases
Group 3 (Subluxation)	
a—Moderate	10
b—Severe	15
Group 4 (Dislocation)	
a—Secondary acetabulum	17
b—No acetabulum	18
Total	60

TABLE III

RESULTS OF UNILATERAL SHELF OPERATION IN SIXTY CASES

Case	Sex	Age at Operation (*Years*)	Group		Follow-up Period		Result of Operation
			Hip Operated Upon	Hip Not Operated Upon	(*Years*)	(*Months*)	
Group 3*a*. Moderate Subluxation							
1	F.	10	3*a*	1*a*	2	8	Good
2	F.	10	3*a*	2*b*	5	3	Good
3	F.	10	3*a*	2*a*	4	6	Good
4	F.	13	3*a*	2*b*	5	3	Fair
5	M.	19	3*a*	1*a*	8	8	Good
6	F.	20	3*a*	3*a*	6	0	Good
7	M.	21	3*a*	2*a*	2	8	Fair
8	F.	30	3*a*	1*a*	3	3	Unimproved
9	M.	35	3*a*	2*a*	4	2	Fair
10	F.	40	3*a*	1*a*	9	5	Good
Group 3*b*. Severe Subluxation							
11	F.	3	3*b*	3*b*	3	6	Unimproved
12	F.	8	3*b*	1*b*	2	4	Good
13	F.	9	3*b*	1*a*	3	3	Unimproved
14	F.	14	3*b*	1*a*	3	9	Good
15	F.	14	3*b*	1*b*	4	5	Unimproved
16	F.	14	3*b*	1*a*	6	9	Good
17	M.	16	3*b*	1*a*	3	0	Unimproved
18	F.	19	3*b*	1*a*	5	9	Good
19	F.	19	3*b*	1*a*	6	9	Good
20	F.	20	3*b*	2*a*	2	11	Good
21	F.	24	3*b*	2*a*	4	11	Unimproved
22	F.	30	3*b*	1*a*	5	6	Good
23	F.	35	3*b*	1*a*	3	11	Good
24	F.	35	3*b*	3*b*	2	5	Unimproved
25	F.	39	3*b*	4*b*	7	1	Unimproved

TABLE III (*Continued*)

Case	Sex	Age at Operation (*Years*)	Group		Follow-up Period		Result of Operation
			Hip Operated Upon	Hip Not Operated Upon	(*Years*)	(*Months*)	
Group 4*a*. Dislocation with Secondary Acetabulum							
26	M.	3	4*a*	1*a*	1	10	Unimproved
27	F.	6	4*a*	1*a*	1	1	Good
28	F.	11	4*a*	2*a*	5	10	Fair
29	F.	12	4*a*	2*b*	13	2	Unimproved
30	M.	14	4*a*	1*a*	4	10	Fair
31	M.	14	4*a*	1*a*	2	3	Good
32	F.	14	4*a*	2*b*	4	0	Fair
33	M.	18	4*a*	2*a*	5	9	Good
34	F.	19	4*a*	3*a*	6	5	Fair
35	F.	19	4*a*	1*a*	3	6	Fair
36	F.	21	4*a*	1*a*	7	6	Good
37	F.	22	4*a*	3*b*	8	3	Fair
38	F.	31	4*a*	1*a*	3	11	Good
39	F.	35	4*a*	4*b*	6	0	Fair
40	F.	36	4*a*	1*a*	4	10	Good
41	F.	36	4*a*	1*b*	2	10	Fair
42	F.	38	4*a*	1*a*	7	2	Good
Group 4*b*. Dislocation. No Secondary Acetabulum							
43	M.	3	4*b*	1*a*	4	8	Unimproved
44	F.	3	4*b*	1*a*	2	10	Good
45	F.	4	4*b*	1*a*	4	4	Unimproved
46	F.	5	4*b*	1*a*	2	4	Unimproved
47	F.	9	4*b*	3*a*	4	2	Unimproved
48	F.	10	4*b*	4*a*	1	7	Unimproved
49	F.	10	4*b*	1*a*	1	4	Good
50	F.	11	4*b*	1*a*	12	1	Good
51	F.	11	4*b*	1*a*	4	5	Unimproved
52	F.	11	4*b*	1*a*	5	9	Good
53	F.	13	4*b*	3*a*	7	11	Good
54	F.	15	4*b*	2*a*	3	2	Good
55	F.	21	4*b*	3*a*	4	4	Unimproved
56	F.	26	4*b*	1*a*	4	8	Fair
57	F.	26	4*b*	1*a*	12	0	Good
58	F.	27	4*b*	1*a*	5	2	Fair
59	F.	29	4*b*	1*a*	1	6	Unimproved
60	F.	31	4*b*	1*a*	9	0	Good

months. It was possible to re-examine forty-three of these patients within the follow-up period. Information was obtained by letter from the other twenty-seven patients.

A completely satisfactory method of classifying the results of the shelf operation is difficult to devise. The authors estimated the patient's degree of functional impairment before and after operation as regards limp, fatigue, pain, and stiffness. If the patient evidenced definite improvement in one or several of these categories and the roentgenograms showed a shelf which adequately covered the head of the femur, the result was classified as "good" or "fair". If the residual disability was slight and did not handicap the individual in his occupation, the result was considered "good". When the residual symptoms

TABLE IV

ANALYSIS OF EIGHTEEN CASES IN WHICH THE PATIENTS WERE UNIMPROVED AFTER UNILATERAL SHELF OPERATION

Case No.	Age (*Years*)	Sex	Group	Remarks
8	30	F.	3*a*	Shelf too high to function well; arthrodesis performed later because of pain in hip
11	3	F.	3*b*	Shelf too high to function well
13	9	F.	3*b*	Small shelf; symptoms not improved
15	14	F.	3*b*	Shelf high; improvement after a subsequent shelf operation
17	16	M.	3*b*	Shelf high; symptoms not improved
21	24	F.	3*b*	Good shelf but aseptic necrosis of head of femur developed; increased pain
24	35	F.	3*b*	Shelf high, questionable function; osteo-arthritis present
25	39	F.	3*b*	Increased pain, narrowed joint space, arthritis; shelf angles upward
26	3	M.	4*a*	Roentgenogram showed head of femur outside shelf
29	12	F.	4*a*	Shelf absorbed
43	3	M.	4*b*	Shelf small and too high to function well
45	4	F.	4*b*	Shelf absorbed
46	5	F.	4*b*	Shelf absorbed
47	9	F.	4*b*	Sciatic neuritis and infected wound postoperatively; hip ankylosed in good position
48	10	F.	4*b*	Spontaneous absorption of head of femur of opposite hip; severe scoliosis; considerable disability
51	11	F.	4*b*	Good shelf; function unimproved
55	21	F.	4*b*	Shelf absorbed
59	29	F.	4*b*	Adequate shelf; no improvement in symptoms

or disability were of moderate degree, the result was considered "fair". If the individual showed no improvement, if the symptoms were worse in any of the categories listed, or if the roentgenograms disclosed a shelf which was too high or too small to cover the head of the femur adequately, the result was classified as "unimproved".

In determining the functional status of the individual, each symptom was graded on the basis of 1 to 4, in which 1 represents a slight degree and 4 a very severe degree of impairment. Absence of the symptom was shown by the figure 0. The following gradations were used:

Limp

Grade 1—slight, may be controlled.
Grade 2—moderate.
Grade 3—severe.
Grade 4—very severe, may require crutches or cane.

Fatigue

Grade 1—hip "feels weak" after strenuous exercise or work.
Grade 2—some "weakness" present daily.
Grade 3—interferes with work.
Grade 4—very severe, requires crutches or frequent rest periods.

Pain

Grade 1—after strenuous exercise, occasional.
Grade 2—after moderate exertion or after day's work.
Grade 3—daily, after mild exertion; interferes with work.
Grade 4—almost constant, incapacitating.

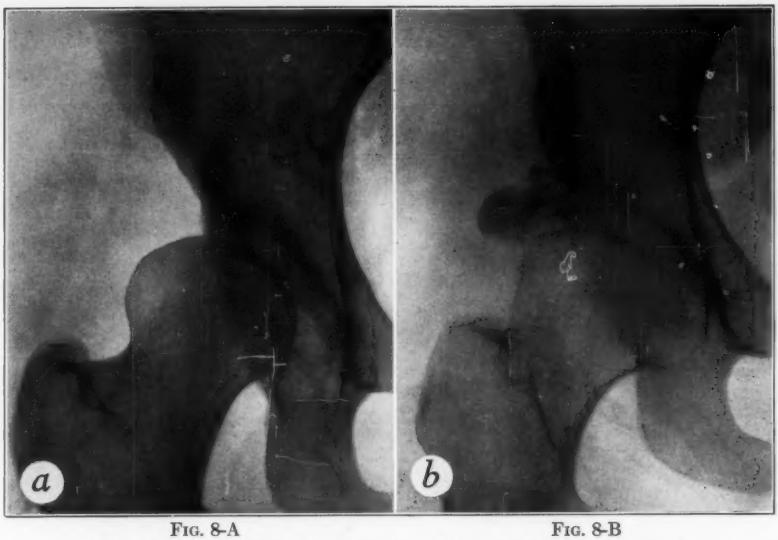

FIG. 8-A FIG. 8-B

Fig. 8-A: Type 3*b*, congenital subluxation of the right hip before operation.
Fig. 8-B: Two weeks after operation.

Stiffness

Grade 1—less than 10 degrees of restriction in any direction; does not limit activity.

Grade 2—at least 90 degrees of flexion and 10 degrees of rotation; able to tie shoes.

Grade 3—less than 90 degrees of flexion; patient may be unable to sit comfortably.

Grade 4—less than 10 degrees of motion in any direction.

In addition, notation was made in each instance of the measured shortening of the affected extremity. The result of the Trendelenburg test was also noted when this information was available.

FIG. 8-C

Three years and three months after operation. There were hypertrophy and adaptation of the shelf, but aseptic necrosis of head of the femur. The condition was considered unimproved.

RESULTS OF UNILATERAL SHELF OPERATION IN SIXTY CASES

The unilateral shelf operation was performed on sixty-six patients. Adequate clinical records of the postoperative course were not available in six instances, and these cases were discarded from the study. Thus, sixty unilateral shelf operations were left for consideration. Fifty-one of the patients were females and nine were males. Their ages ranged from three to forty years, the average being eighteen years. The hips operated upon were classified on the basis of their roentgenographic features (Table II).

In evaluating the results of these operations, the clinical records and the roentgenograms taken before and after operation were compared. The disability was evaluated in terms of limp, fatigue, pain, and decrease in range of motion in the hip operated upon, as described previously. Thirty-five patients returned for re-examination during the follow-up

TABLE V

PREOPERATIVE ANATOMICAL CLASSIFICATION OF HIPS OF TEN INDIVIDUALS ON WHOM BILATERAL SHELF OPERATION WAS PERFORMED

Classification	No. of Hips
Group 3 (Subluxation)	
a—Moderate	2
b—Severe	4
Group 4 (Dislocation)	
a—Secondary acetabulum	6
b—No acetabulum	8
Total	20

period. Information upon which to judge the operative results was secured by letter from the remaining twenty-five patients.

TABLE VI

RESULTS OF BILATERAL SHELF OPERATION IN TEN CASES

Case No.	Sex	Age at Operation (*Years*)	Hip Operated Upon	Group	Follow-up Period (*Years*)	(*Months*)	Result of Operation
61	F.	18	Right	3*b*	3	7	Good
			Left	3*b*	3	7	Good
62	F.	23	Left	3*b*	4	0	Fair
			Right	3*a*	3	11	Fair
63	F.	40	Right	3*a*	4	10	Good
			Left	3*b*	4	9	Fair
64	F.	7	Right	4*b*	8	6	Fair
			Left	4*b*	7	9	Good
65	M.	8	Right	4*b*	3	10	Good
			Left	4*a*	3	3	Good
66	F.	9	Left	4*b*	4	9	Unimproved
			Right	4*a*	4	7	Good
67	F.	13	Right	4*a*	2	4	Good
			Left	4*b*	2	3	Good
68	M.	16	Right	4*a*	6	8	Unimproved
			Left	4*a*	6	8	Good
69	F.	23	Left	4*b*	1	1	Unimproved
			Right	4*b*	1	0	Unimproved
70	F.	30	Left	4*a*	7	9	Fair
			Right	4*b*	7	7	Good

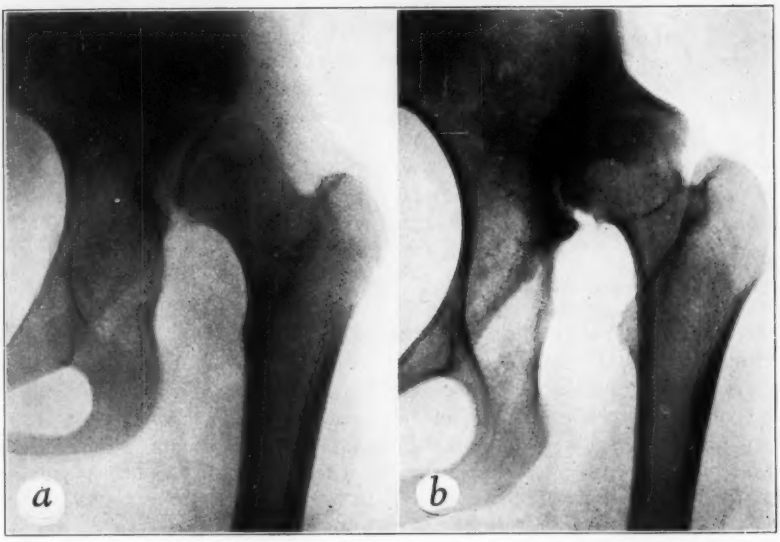

FIG. 9-A FIG. 9-B

Fig. 9-A: Group 4*a*, congenital dislocation of the left hip before operation.
Fig. 9-B: Five years and ten months after a shelf operation over the false acetabulum, the result was fair.

The final results of this operative procedure were good in twenty-nine cases, fair in thirteen, and unimproved in eighteen (Table III). The clinical records and roentgenograms of the eighteen patients who failed to show improvement were reviewed to determine whether or not any explanation could be found for the unsatisfactory outcome (Table IV).

RESULTS OF BILATERAL SHELF OPERATION IN TEN CASES

Shelves were constructed over both hips in ten patients,—eight females and two males. Their ages at the time of operation ranged from seven to forty years, the average being eighteen years. These hips were classified on the basis of their roentgenographic features before operation (Table V).

Adequate follow-up records were available for all ten patients in this group. The duration of the follow-up period ranged from one year to eight years and six months, with an average of four years and two months. It was possible to re-examine eight patients during the follow-up period. Information was obtained by letter from the other two patients. The result was good in eleven hips, fair in five hips, and unimproved in four (Table VI).

The reasons for the unsatisfactory results obtained in four hips are given in Table VII.

TABLE VII

ANALYSIS OF DATA ON FOUR HIPS NOT IMPROVED AFTER BILATERAL SHELF OPERATION

Case No.	Age (*Years*)	Sex	Hip	Group	Remarks
66	9	F.	Left	4*b*	Shelf absorbed; improvement after another shelf operation
68	16	M.	Right	4*a*	Ankylosis; absorption of head of femur
69	23	F.	Left	4*b*	New acetabula and shelves were fashioned in walls of ilium; symptoms not improved; improvement later after bilateral cup arthroplasty
			Right	4*b*	

COMMENT

The benefit derived by an individual from any surgical procedure may be judged in terms of relief of symptoms or deformity, improved function, and prevention of disabling sequelae. While it is difficult to state what the future course would have been for any individual in this series had the shelf operation not been performed, we believe that this operation prevents or delays the onset of the arthritic changes which accompany disordered joint function. In the younger individuals, limp and fatigue were the most prominent symptoms. In the older patients, pain, fatigue, and limitation of motion were more frequent complaints.

Limp

A limp was present in almost all of the patients in this series, but many, especially the younger individuals, were capable of a considerable amount of exertion despite a severe limp. The limp depends upon shortening and deformity of the femur, absence of bone support for the pelvis upon the femoral head, weakness of the hip abductors (Trendelenburg phenomenon), and the presence of pain. If the shortening is overcome by traction and is maintained by a well placed shelf, if the hip abductors are strengthened by exercises, or if the pain is relieved after the operation, the limp may be improved (Figs. 3-A and 3-B). In most of the cases, the patient continued to limp after operation. In forty-four patients, it was thought that the limp decreased after the shelf operation; in three patients the limp may have been more pronounced. This does not reflect in a derogatory manner upon the operation as performed, because in most instances it was not possible to restore the head of the femur to its normal position, and the shortness of the extremity remained to produce a limp (Figs. 9-A and 9-B). Overambitious attempts to restore length to the extremity by the use of heavy traction may result in sciatic neuritis, weakening of the hip abductors through overstretching, and disruption of the shelf, if the traction is not maintained efficiently during the prolonged convalescent period while the shelf is consolidating.

Lordosis

In complete bilateral dislocation of the hip, increased lumbar lordosis usually develops because the patient attempts to center his weight over the posteriorly displaced heads of the femora. The attempt was usually made at operation to place the posteriorly dislocated heads of the femora into a more anterior position to improve the patient's posture. None of the patients in this series complained of backache which might be attributed to increased lumbar lordosis.

Pain

Pain was usually well localized about the hip, although a few patients experienced pain referred to the knee. Forty-eight of the seventy patients complained of pain before operation. The average age at onset of this symptom was twenty-one years. There was a striking absence of this symptom among individuals under the age of eleven and among those whose hips were completely dislocated without the development of a secondary acetabulum.

After operation, eighteen patients were completely relieved of pain and twenty had less pain. Ten patients noticed no difference in the amount of pain and two patients experienced pain in the hip where they had none prior to operation.

Fatigue

Patients frequently complained that a hip "felt weak" or that their endurance was low. Fatigue was an indefinite type of complaint which was usually present; it was difficult

TABLE VIII

RESULTS OF EIGHTY SHELF OPERATIONS

Operation	No. of Hips	Result		
		Good	Fair	Unimproved
Unilateral shelf	60	29	13	18
Bilateral shelf (10 patients)	20	11	5	4
Total	80	40	18	22

to evaluate, and varied in degree with the amount of pain and limp. The authors estimated that fatigue was decreased following the shelf operation in thirty-seven patients, unchanged in twenty-four, and increased in two. This complaint could not be evaluated in the remainder.

Stiffness

Motion in the hips operated upon was evaluated in terms of restriction of the normal range. Limitation of motion did not necessarily restrict a patient's activity and did not constitute a serious handicap unless severe. Some limitation of motion was noted in the affected hip of forty-eight patients before operation. The records indicated that in eighteen patients there was no appreciable limitation of motion following the shelf operation; in twenty-eight the range of motion was the same after operation as before; in fourteen the range was decreased and in ten the range was increased after the shelf operation.

The results of eighty shelf operations, performed as a unilateral or bilateral procedure on seventy patients with congenital subluxation or dislocation of the hip, are given in Table VIII.

Failure of the operation in those cases classified as unimproved was attributed in seven cases to a shelf which was too small or poorly placed (Figs. 7-A and 7-B). Absorption of the shelf was noted in five cases. In two cases, the hip became ankylosed after operation. In one case, aseptic necrosis occurred in the head of the femur on the side operated upon (Figs. 8-A, 8-B, and 8-C). In several instances, the symptoms which persisted after operation could have been attributed to arthritic changes within the joint. One patient had transient sciatic neuritis following heavy traction and the construction of a shelf.

SUMMARY

Although the exact cause of congenital dysplasia of the hip is unknown, genetic and biomechanical factors are probably important in producing the malformation, which may be hereditary and frequently involves both hips in varying degrees. Determination of the CE angle of Wiberg is helpful in classifying the various degrees of subluxation of the hips, as revealed by roentgenogram.

The purpose of the shelf operation is to increase the weight-bearing surface of the malformed acetabulum and thus to afford stability for the subluxated or dislocated femoral head. The operation consists in construction of an adequate bone ledge or shelf on the side wall of the ilium over the head of the femur, after the femur has been displaced downward by traction (Figs. 2-A and 2-B).

In a series of eighty operations for congenital subluxation or dislocation of one or both hips, fifty-eight (72.5 per cent.) were successful in producing partial or entire relief of limp, pain, and hip fatigue, while a good range of motion was retained in the hip operated upon. Twenty-two of the operations (27.5 per cent.) were unsuccessful, in that the patients' symptoms were unimproved. Poor results were attributed to construction of shelves which

were inaccurately placed, were too small, or were absorbed. Arthritic symptoms persisted or developed after operation in a few instances.

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DISCUSSION

The Use of a Metallic Femoral Head

(*Continued from page 75*)

be done on a wholesale basis until much more is learned about it. I think the use of the operation should be restricted to a small group of men who can do it experimentally for a time. Dr. Hudack showed a number of his results to us last year and, functionally, they were poor. These patients walked badly, and in three cases the hips became dislocated. Each speaker has mentioned a number of cases in which the prosthesis has become displaced, so that is another complication which must be looked for.

Dr. Peterson (closing): Dr. Moore has asked me to comment on the presence of pain in these hips. It would hardly be possible to say that none of these patients has pain after either ankylosis, arthritis, or non-union with prolonged disability. However, I feel that their pain has been minimal in degree and of as mild a nature as might be expected after the pre-existing illness or disease and one or more reconstructive procedures.

In regard to movement, the metal parts are very accurately fitted together and are difficult to separate after the prosthesis has been assembled. After further impaction by weight-bearing, motion between these parts is minimal or non-existent.

Reference has been made to the type of metal. There are many types of stainless steel and all are not physiologically inert. However, careful research has shown that this type of steel (18-8 SMo) is comparable to Vitallium in its inertness and superior in its strength and physical qualities.

The magnitude of the surgery should not be underestimated, but it is comparable to other reconstructive procedures on the hip. You will note that, of approximately 100 reconstructions of this type to date, done by about twenty surgeons in scattered areas of this country, there has been no operative mortality, which is a good record. The precautions stated by the discussors are entirely correct. There will no doubt be further complications because of the weakness of the earlier devices, but I am confident that some of the unfavorable prophecies made today will not come true.

LOCALIZED NEUROFIBROMATOSIS WITH ASSOCIATED OVERGROWTH OF AN EXTREMITY

Equalization of Limb Length Following Epiphyseal Arrest

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Marked inequality of limb length, caused by a variety of pathological processes, has always presented a difficult corrective problem for the orthopaedic surgeon. The occurrence of overgrowth of an extremity, associated with neurofibromatosis—either localized to the involved extremity or generalized neurofibromatosis (von Recklinghausen's disease)—has apparently received detailed attention in the literature only in the past decade.

Generalized neurofibromatosis was first described by von Recklinghausen in 1882. Cases which are now recognized as neurofibromatosis were reported as early as 1842, but bone changes in this condition escaped detection until early in this century. Various authors [5, 6] reported the similarity of the subperiosteal bone cysts occurring in generalized neurofibromatosis to the bone cysts observed in osteitis fibrosa cystica. However, it is now agreed that the pathogenesis of these two conditions is entirely different. Variation in terminology is still a confusing feature. Lichtenstein suggested "polyostotic fibrous dysplasia" for bone changes occurring in neurofibromatosis, while Albright and Reifenstein preferred "osteitis fibrosa disseminata". Brooks and Lehman classified the various osseous changes in neurofibromatosis as: (1) scoliosis (by far the most common); (2) abnormalities of growth (most commonly associated with elephantiasis); and (3) subperiosteal bone cysts.

Moore reported four cases of localized hypertrophy of an extremity associated with neurofibromatosis, in which the neurofibromatosis was either local or generalized. He disagreed with the then accepted idea that the hypertrophy was caused by bony invasion of the neurofibromatous tissue, and offered instead a logical belief: "The hypertrophy is not pure overgrowth, but is mixed over and underdevelopment, and we believe represents uncontrolled growth rather than stimulation of growth". In contrast, Friedman reported four cases with actual bone invasion by neurofibromatous tissue and resultant marked hypertrophy and deformity of the involved bone. Uhlmann and Grossman reported one case of invasion of the mandible by neurofibromatous tissue with enlargement of the jaw. These authors included one additional case of unilateral enlargement of the leg secondary to soft-tissue neurofibromatosis. It was their feeling that biopsy was hazardous, because of the danger of resultant malignant metaplasia. They did not recommend epiphyseal arrest of the affected limb, because of the necessity of cutting through the neurofibromatous tissue. Malignant degeneration occurring in neurofibromatosis has been reported by Sharpe and Young.

The occurrence of pseudarthrosis in association with generalized neurofibromatosis is well recognized. Barber, in 1939, called attention to the presence of bowing of the leg with neurofibromatosis, and pointed out that apparently many similar cases have been classified as congenital pseudarthrosis. He felt that neurofibromatosis might have escaped detection in many of these cases. Jacobs, Kimmelstiel, and Thompson, among others, pointed out the association of pseudarthrosis with neurofibromatosis. Some authors [4, 13] believe that congenital pseudarthrosis occurs only in patients having generalized neurofibromatosis.

Green and Rudo reported a case of pseudarthrosis of the tibia and fibula and included

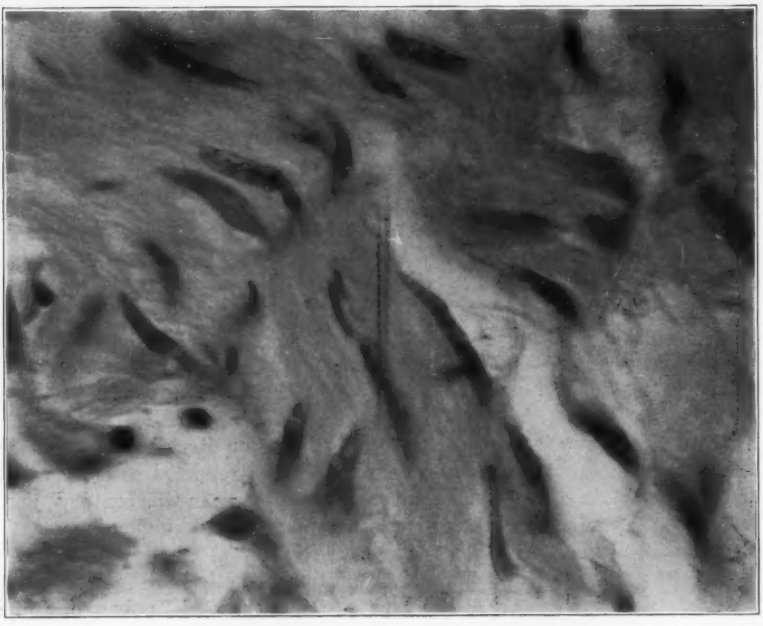

FIG. 2

Photomicrograph of biopsy material. Note whirls and palisading of nuclei.

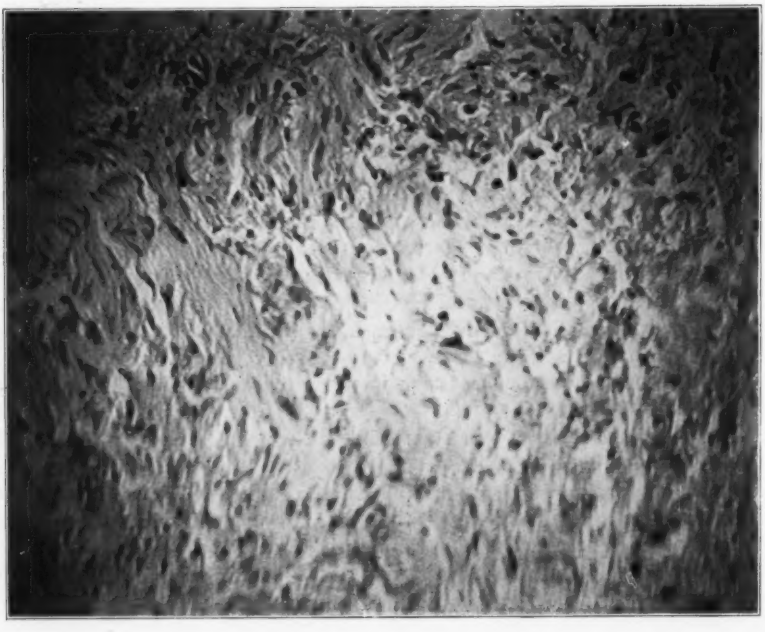

FIG. 1

Photomicrograph (× 240) of biopsy specimen of neurofibroma removed at time of original exploration.

photomicrographs of tissue removed from the fibula at the site of the pseudarthrosis. This tissue showed the nuclear palisading and swirling with interlacing bundles, characteristic of neurofibromatosis. This evidence suggests that there is actually bone invasion by the pathological tissue. In confirmation of this, Gross, Bailey, and Jacox reported a case of intramedullary neurofibroma of the shaft of the humerus, which showed the characteristic histological picture of neurofibromatosis. There was no evidence of generalized neurofibromatosis in this patient.

The following case is reported because of the unusual occurrence of a localized neurofibroma, which apparently started to grow rapidly several months after relatively minor trauma. There was marked overgrowth of the affected limb in a year. Evidence of generalized neurofibromatosis was not present in this patient, and there was no history of such a lesion in any member of her family.

L. D. was first seen in June 1946, at the age of ten years, for treatment of a sprain of her right ankle. This healed without sequelae and she was not seen again until April 1947, when she complained of swelling and tenderness of the right ankle. A diffuse soft-tissue swelling over the medial aspect of the ankle was tender to pressure and caused radiation of pain into the plantar surface of the foot. Roentgenograms of the foot and ankle showed no evidence of bone abnormality. Because of a slight increase in the size of the soft-tissue mass, it was explored in June 1947. At operation the lesion was found to arise from the tibial nerve; it extended, with many ramifications, under the sole and along the medial aspect of the foot. It also extended superiorly into the calf, following the course of the tibial nerve. A neurosurgeon, consulted at the time of operation, felt that complete excision of the mass would injure the nerve supply to the foot and would cause extensive disfigurement of the limb. The wound was closed after partial resection of a considerable area of the tumor. Sections from this tissue were reported by two pathologists as presenting the histological picture of benign neurofibroma (Figs. 1 and 2).

Both lower extremities were equal in length and diameter at that time. The patient was observed at frequent intervals following operation and no evidence of increase in size of the mass was noted. Approximately eleven months later, when she was twelve years of age, the right leg was one and one-half inches longer than the left; most of the increase in growth was in the tibia.

FIG. 3

Left knee. Comparison roentgenogram, five months after epiphyseal arrest of right tibia, fibula, and femur.

FIG. 4

Right knee. Five months after arrest of distal femoral epiphysis and proximal epiphyses of tibia and fibula.

FIG. 5

Right knee. Seventeen months after epiphyseal arrest, epiphyseal lines are almost obliterated.

Epiphyseal arrest of the distal femoral and proximal tibial epiphyses on the right was recommended, in order to equalize the limb lengths. The patient's family obtained further consultation with orthopaedic surgeons. Both of these consultants agreed with the recommendation of epiphyseal arrest.

This procedure was carried out in July 1948 by curettage of the epiphyseal plates of the distal femoral and proximal tibial epiphyses, and the proximal ephiphysis of the fibula. At the time of operation, the right lower extremity was one and one-half inches longer than the left. Five months after operation, it was one inch longer than the left (Figs. 3 and 4); and ten months after operation the left leg was only one-half inch short. Seventeen months postoperatively (Fig. 5), the difference in limb lengths was only one-quarter of an inch. Roentgenograms at that time showed that the arrested epiphyseal plates were completely closed; the corresponding epiphyseal plates on the left were very slightly visible, suggesting that equalization of limb length would be obtained at the time of cessation of bone growth.

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THE SURGICAL MANAGEMENT OF CHRONIC OSTEOMYELITIS BY SAUCERIZATION, PRIMARY CLOSURE, AND ANTIBIOTIC CONTROL

Preliminary Report on Use of Aureomycin*

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The advent of the antibiotics has brought new hope to the age-old quest [1-3, 5-11, 15] for more effective treatment of chronic osteomyelitis. The flush of early successes has led to claims [4, 12-14, 17] which may now be subjected to an intermediate evaluation in the light of additional observations and studies. In 1945, in a preliminary report on "Penicillin in the Treatment of Chronic Osteomyelitis", the authors [16] referred to a clinical and bacteriological study carried out under the aegis of the Committee on Medical Research of the Office of Scientific Research and Development at the Hospital for Joint Diseases. The results were detailed for the treatment of chronic osteomyelitis with penicillin, with and without surgical intervention, in forty-seven cases, with a total of sixty-eight lesions. The conclusions were:

1. The treatment of chronic osteomyelitis with parenteral and local applications of penicillin, without surgical intervention, was totally ineffective. Not a single osseous lesion was improved by this method. These failures were thought to be due to the inherent characteristics of the lesion—namely, its greatly impoverished blood supply and the complete avascularity of the contents of the bone cavities, such as pus and sequestra—resulting in the inability of the blood stream to deliver the antibiotic to the focus of infection.

2. The treatment of chronic osteomyelitis with preoperative and postoperative intramuscular penicillin, thorough saucerization of the focus, and topical application of penicillin was not materially influenced by the antibiotic, because the healing time by secondary intention was not appreciably shortened.

3. The treatment of chronic osteomyelitis with preoperative and postoperative intramuscular penicillin, thorough saucerization of the focus, topical application of penicillin, and delayed primary closure of the resultant wound was a considerable improvement over previous methods, because of the appreciable shortening of the healing time by delayed primary intention.

4. The treatment of chronic osteomyelitis with preoperative and postoperative intramuscular penicillin, thorough saucerization of the focus, local application of penicillin, and primary closure of the resultant wound was an extraordinary improvement on previous methods, because twenty-three of twenty-seven lesions healed by primary intention.

5. The presence of secondary organisms prolonged the time of healing of the saucerized areas. A distinct advantage of treatment by primary closure is the reduction of the opportunity for contamination of the wound by "nuisance" organisms, particularly *Bacillus proteus* and *Bacillus pyocyaneus*. Such organisms were invariably present in wounds allowed to heal by secondary intention; they were present in 80 per cent. of those wounds allowed to heal by delayed first intention subsequent to delayed closure; and were present in only five of twenty-seven wounds allowed to heal by first intention subsequent to primary closure without drainage.

6. Staphylococci which persisted over long periods of time in wounds healing by secondary or delayed primary intention tended to develop increasing resistance to peni-

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cillin. In only one of the patients treated by primary closure was the development of resistance of the organisms noted. In no instance did increased resistance of Staphylococci occur in patients treated by penicillin without surgical intervention. The material for cultures from such patients was necessarily obtained from draining sinus. It is, therefore, probable that such organisms were not exposed to penicillin administered intramuscularly, because of the relatively avascular nature of the thickened fibrous walls of the sinus and the avascularity of the lesions in which they originated.

Subsequent to these findings, the authors' project, devoted to the treatment of chronic osteomyelitis by saucerization and primary closure under antibiotic control, was continued at the Hospital for Joint Diseases. Our present management of a case of chronic osteomyelitis begins with a thorough review of the history with special reference to previous operative procedures, previous administration of antibiotic therapy, and previous occurrences of allergic and toxic reactions to the agent. In the physical examination, special note is made of the availability of good skin coverage subsequent to the operative intervention. Roentgenographic examinations include, in addition to flat views, stereoscopic studies made subsequent to the injection of lipiodol for the visualization of the course of sinus tracts and the localization of cavities in difficult areas, such as the pelvis. On occasion, laminographic studies are made. Immediately after the patient is admitted and before the sinus are injected for roentgenographic studies, cultures are taken and the sensitivities of the isolated organisms to the various antibiotics are determined. Blood studies are made to determine the presence of anaemia, which is treated appropriately with medications and blood transfusions prior to, during, and after the operative procedures.

The therapy of the osteomyelitic lesion usually begins on the day before operative intervention with the intramuscular administration of an aqueous solution of penicillin, which is continued thereafter every three hours for a minimum period of two weeks. Studies have shown that 20,000 units administered every three hours gives sufficient blood levels in about 80 per cent. of the osteomyelitic lesions in which the organisms are sensitive to 0.2 unit of penicillin. Because of the increased availability of the drug, the more frequent loss of sensitivity of the organism to penicillin, and the absence of bacteriological data in those cases which have no discharging wounds, the dosage has been gradually increased, so that at the present time we administer 50,000 units of penicillin intramuscularly every three hours. In the presence of micro-organisms resistant to 0.2 unit or more of penicillin, larger doses are administered. At times streptomycin is added to the penicillin in the event that the organism is resistant to penicillin and sensitive to streptomycin. A usual dose of streptomycin at the present time is 0.15 gram every three hours. Larger doses are used in resistant cases. The use of aureomycin is recent and will be discussed later.

The surgical procedure is carried out with the patient under general anaesthesia and, wherever feasible, under tourniquet control; the tourniquet is not released until after closure of the wound and the application of a compression dressing of sheet wadding, flannel bandage, and adhesive. The operative incision is sufficiently extensive to expose normal bone at the extremities of the lesion and to allow the most direct approach to the diseased area without endangering important blood vessels, nerves, or muscle mechanisms. The soft-tissue scar is excised to the greatest extent possible. The periosteum and soft tissues are stripped only from those areas of bone to be excised. Long bones are usually unroofed with a motor saw, and the lesion is saucerized with S-shaped gouges and chisels, normal bone and medullary canal being exposed at the extremities of the lesion. The walls of the saucerized area are leveled off to make a shallow cavity; all visible foci and overhanging ledges are removed. The wound is then thoroughly cleansed and flooded with a solution of saline containing 250 units of penicillin per cubic centimeter. In those instances which present a mixed infection or resistant organisms, the penicillin content of the solution is raised and 1,000 micrograms of streptomycin per cubic centimeter are

added. The wound is then closed without drainage, in several layers wherever possible. When there is insufficient skin for a satisfactory closure, a dermoplastic procedure is carried out by undermining the skin and utilizing releasing incisions; these incisions may or may not be closed, depending upon the local conditions. Sinus tracts are excised wherever possible. If excision is not feasible, the tracts are curetted and the openings are closed with one or two sutures. Wet dressings containing the antibiotic solution are applied after skin closure with silk. A compression bandage of sheet wadding, flannel, and adhesive is used to immobilize the limb and to support its circulation. Plaster-of-Paris support is used only when an excessive amount of bone has been removed and postoperative fracture is feared.

The wound is left undisturbed until the tenth postoperative day, at which time the sutures are removed. In the great majority of instances, healing by primary intention is found at this time. The line of incision is nevertheless supported with adhesive bridges, because fluctuation is still evident in the superficially closed wound. It is most important that the underlying hematoma be not discharged. Evacuation of the hematoma results in the formation of a cavity which can only be healed by granulations, thus defeating the objective of healing by primary intention. In the event that partial evacuation of the hematoma occurs, the wound is covered with a dry dressing and the limb is further immobilized in a compression dressing. At no time is the wound probed, drained, or irrigated. Healing occurs, although it may be delayed for several weeks.

This intermediate report presents a series of 100 saucerization procedures with primary closure of the resultant wounds under antibiotic control in ninety-three lesions, occurring in eighty-three patients. All procedures carried out under aureomycin control will be dealt with separately, later in this presentation. The ninety-three lesions include some previously reported and represent work done during the past five and one-half years. Several of these patients have been observed recently, while others have been followed for varying periods of time up to five and one-half years. Sufficiently frequent follow-up contacts have been maintained in a large proportion of instances to make this statistical study reliable.

The mere demonstration of percentile data would present a very inadequate picture of the results obtained unless there be included a sufficiently detailed description of the character of the cases involved. Such a detailed study of ninety-three lesions would be impracticable because of lack of time and space. Many of the patients presented multiple and extensive lesions of long duration. Some had single or multiple sinus of long standing, while others had no draining lesions. A detailed analysis of these factors would aid in evaluating the results shown in this report; this is, nevertheless, impractical and will not be undertaken. A detailed consideration of previous therapy, although of the utmost importance, cannot be undertaken at this time. However, practically all of these patients have had many previous operations carried out according to the more or less standardized techniques, such as the Carrel-Dakin method, Orr technique, use of Baer's maggots, or other methods. Some have had previous antibiotic therapy during the acute, post-acute, or chronic stages with or without surgical intervention. It is thus evident that there are many elements in this problem which cannot possibly be included in a report such as this. Some of the high lights are presented in the hope of inducing additional study.

In Table I are presented data showing the distribution of patients according to age groups; the proportion of multiple and solitary osteomyelitic lesions; the longest, shortest, and average duration of the disease in each age group; the number of operations performed; the number of lesions healed *per primam;* and the number of recurrences. The lesions are classified according to the basis of origin,—that is, hematogenous or exogenous. In the exogenous group are included all osteomyelitic lesions which arose subsequent to compound fractures, gunshot wounds, postoperative bone infections, and bone infections by contiguity.

TABLE I

CLINICAL DATA IN EIGHTY-THREE PATIENTS

Age Groups (*Years*)	Total Patients	Patients with Multiple Lesions	Patients with Solitary Lesions	Duration of Disease			No. of Operations	Healed *per Primum*	Recurrences	Origin of Disease	
				Longest (*Years*)	Shortest	Average (*Years*)				Hematogenous	Exogenous
0 to 10	4	1	3	2	4 mos.	1+	5	5		5	0
11 to 20	14	5	9	9	1 mo.	3+	21	17		20	1
21 to 30	24	6	18	24	2 mos.	12−	28	19	2	21	7
31 to 40	16	4	12	32	3 mos.	14−	16	16	1	11	5
41 to 50	13	0	13	37	3 mos.	22+	17	9		11	6
51 to 60	10	2	8	43	3 mos.	17	11	11		5	6
69 and 72	2	0	2	62	8 yrs.		2	1			2
Total	83	18	65				100	78	3	73	27

Table II shows that of ninety-three lesions of chronic osteomyelitis treated in eighty-three patients, 73 (79 per cent.) healed by primary or delayed primary intention. Under the term "healing by delayed primary intention" are included those instances in which, at the time of removal of sutures, some oozing was present due to spontaneous partial evacuation of the underlying hematoma. This in a few instances persisted up to six weeks. One lesion (1 per cent.) involved a lower jaw, and what appeared to be a complete loss of the hematoma occurred with drainage of the resultant cavity for eight months. Other complicating factors in this instance may have been the inadequate concentration of penicillin in the blood stream during periods of oral administration of the antibiotic, and the use of gelfoam and thrombin locally [18] in an effort to obliterate the cavity resulting from the operative procedure. In other instances of this group of cases, the wound edges presented small areas of skin necrosis due to embarrassed circulation; these were incidental to poor skin coverage or dermoplastic procedures utilized to get primary closure at the time of the saucerization. These lesions usually healed within three to four weeks; a few took as long as four to six weeks. In the greater portion of this group of seventy-three cases, the lesions were completely healed on the day the sutures were removed. In the remainder, healing was by so-called delayed primary intention.

The next group of five lesions (5 per cent.) presented in part more extensive areas of skin necrosis, while the remainder of the wounds were well healed at the time the sutures were removed. These necessitated pinch grafts or dermoplastic procedures with resultant delay in healing to two months in two instances, and to three, four, or five months in the remaining instances.

In five instances (5 per cent.), failure of healing after the first saucerization was due to inadequate surgical procedures or the presence of resistant micro-organisms. The details follow. In all of these instances, healing was by primary intention subsequent to resaucerization and primary closure.

CASE 24–34. A girl, fourteen years old, who had had hematogenous osteomyelitis of the lower end of the femur of one year's duration, three previous operations on the femur and one arthrotomy, roentgenographic evidence of extensive bone changes, and two persistently draining sinus had been submitted to a saucerization operation, curettage of the sinus tracts, and primary closure under antibiotic control. Despite the administration of over 3,600,000 units of penicillin, the wound broke down repeatedly. Failure was attributed to the presence of a mixed infection and to an inadequate surgical procedure.

A second saucerization was performed five months later (April 9, 1946). The disease was found to extend to the articular cortex of the condyles; the sinus tract led into the suprapatellar pouch. The fibrous scar was extensively excised; the knee joint was exposed during the excision of the sinus tract and a large amount of pus was released. The wound was then closed without drainage. Firm healing was found thirteen days

after operation, and the lesion has remained healed to date. Throughout the entire course, culture of the wound showed penicillin-sensitive, coagulase-positive *Staphylococcus aureus* and penicillin-resistant secondary invaders.

CASE 53–78. A man, twenty-nine years old, gave a history of multiple hematogenous osteomyelitic lesions involving the right humerus, the left forearm, and two foci in the left femur of fifteen years' duration. The femur had drained on and off and had been subjected to twenty operations. Cultures revealed penicillin-sensitive, coagulase-positive *Staphylococcus aureus.*

On February 27, 1945, under antibiotic control, the lower half of the femur was saucerized, the sinus tract was excised, and the wounds were closed without drainage. Eleven days later penicillin had to be discontinued because of severe urticaria and angioneurotic oedema involving the face, mouth, and other parts of the body. After the subsidence of these phenomena, the administration of penicillin was resumed, but the wound broke down and the temperature rose. A total of 3,005,000 units of penicillin was administered. Failure was attributed to premature withdrawal of the antibiotic and an incomplete saucerization procedure.

On April 27, a second operation was performed through a fifteen-inch incision, and the entire femur was resaucerized under antibiotic control. The penicillin dose was raised to 30,000 units every three hours. The wound was found to be healed on the twelfth day and it has remained healed.

CASE 87–117. A man, forty-two years old, had had hematogenous osteomyelitis of the femur since 1915, when he was twelve years of age. From then until 1929, he had five operations. At no time was the wound closed for longer than one and one-half years, and since 1929 it had drained persistently. On August 28, 1945, a saucerization, excision of the sinus tract, and primary closure were performed under antibiotic control. The temperature rose to 103 degrees. When the wound was opened, considerable slough and foul discharge were encountered. Failure was attributed to the presence of *Bacillus proteus* as the predominating organism. Finally a coagulase-positive *Staphylococcus aureus* was isolated, which was found to be resistant to 5 units of penicillin and 25 milligrams of sulfathiazole and sulfadiazine.

On January 14, 1946, the patient was placed on a regimen of 30,000 units of penicillin and 1 gram of sulfathiazole; on the following day the area was resaucerized, with excision of the old scar. Primary closure was performed after the topical application of penicillin and sulfanilamide. Two weeks later the temperature rose to 103 degrees and a profuse discharge occurred. The fever then subsided; the wound healed by delayed primary intention in two months and has remained healed. A total of 6,310,000 units of penicillin and 80 grams of sulfathiazole was administered during the second period of treatment.

CASE 124–178. An eighteen-year-old male had had an osteomyelitic lesion of hematogenous origin for two and one-half years. He presented a draining sinus over the right sacro-iliac area and another just below the anterior-superior spine of the pelvis on the same side, in addition to multiple scars resulting from healed sinus and two operative procedures. Roentgenograms revealed sequestrum formation and extensive involvement of the posterior two thirds of the ilium, extending from the sciatic notch to one inch below the iliac crest. The sinus tracts in front and back communicated with the lesion. The condition was complicated by an acute gonorrheal infection.

On March 26, 1946, subtotal resection of the ilium was performed, the crest of the ilium being left attached to the soft tissues. A hemorrhage of the superior gluteal artery was controlled by packing with oxycel. The wound was closed without drainage. Antibiotic control consisted of the administration of 40,000 units of penicillin every three hours, in addition to topical application at the time of the operation. The wound failed to heal, despite the administration of 6,940,000 units of penicillin. Failure was attributed to inadequate surgery, because roentgenograms showed sequestration of the crest of the ilium and the presence of a penicillin-resistant (to 12 units), coagulase-positive *Staphylococcus aureus.*

On June 7, 1946, another saucerization operation was carried out. The crest of the ilium was removed and primary closure was performed under antibiotic control, 30,000 units of penicillin being administered every three hours. The wound was found to be healed on the eighteenth day and had remained so at the last follow-up contact.

CASE 213–304. A man, forty-five years old, stated that in 1922, six months after an open operation and fixation with a Lane plate for a fracture of the femur, the plate broke. At a second operation a beef-bone graft and a new metal plate were applied. The knee was thereafter manipulated because of loss of motion, resulting in fracture, separation of the fragments, and non-union of the patella; these symptoms have persisted to date. In 1926 a sinus developed, which persisted until 1937; at that time the plate and remains of the graft were removed, and the femur was saucerized and treated according to the Orr technique. The sinus persisted and, on December 31, 1946, the area was saucerized without firm closure of the wound. In spite of antibiotic control, the wound failed to heal.

Thereafter, the patient came under the authors' care. On February 18, 1947, the wound was resaucerized and closed without drainage after the topical administration of penicillin, 500 units per cubic centi-

meter, and streptomycin, 200 micrograms per cubic centimeter. The wound healed, but one week later broke down, despite the intramuscular injection of 30,000 units of penicillin every three hours. Cultures taken at and prior to the operation revealed a mixed infection, consisting of coagulase-positive *Staphylococcus aureus*, sensitive to 0.156 unit of penicillin and resistant to 25 micrograms of streptomycin; *Bacillus pyocyaneus*, which was resistant to 50 micrograms of streptomycin; and secondary invaders. This failure was attributed to the presence of a mixed infection and an extensive soft-tissue avascular scar which could not be sufficiently excised at operation.

Seven months later (September 18) the area was again saucerized, and much of the soft-tissue scar was excised. The wound was closed without drainage under antibiotic control,—50,000 units of penicillin every three hours. Delayed primary healing occurred at the end of seven weeks. The wound has remained healed.

The last five lesions (5 per cent.) in Table II failed to heal after one or more saucerization procedures, either because of resistant organisms, inadequate surgery, or a combination of these factors. In many of these cases, involvement was so extensive as to make surgical intervention dubious. Surgery was nevertheless undertaken with the hope that satisfactory results might be obtained. The details of these cases are as follows:

Case 94–129. A fourteen-year-old male had had acute osteomyelitis of the tibia for six days. He was treated with 20,000 units of penicillin intramuscularly every three hours. Penicillin was injected locally after the aspiration of pus on repeated occasions. During the first two weeks of observation, the patient received 2,340,000 units of penicillin intramuscularly and 76,250 units locally. The systemic symptoms subsided, but the local lesion steadily progressed clinically and roentgenographically. It persistently yielded penicillin-sensitive (0.156 unit), coagulase-positive *Staphylococcus aureus*, notwithstanding the fact that the blood culture became sterile.

One month later (September 25, 1945) the tibia was saucerized and closed without drainage under antibiotic control. Penicillin was discontinued on the twentieth day after operation, at which time the wound was healed. Seven days later the temperature rose and the wound discharged serous material which was sterile on culture. Penicillin therapy was reinstituted for six days, during which time the wound healed. A total of 4,685,000 units of penicillin was administered in conjunction with the operative procedure. The patient was discharged to his home two months after the saucerization.

Three weeks later the lesion recurred and the wound broke down. Penicillin was again administered, and in two weeks the sinus had closed. Four months later the lesion broke down again, and roentgenographic studies suggested sequestrum formation. The wound healed after several weeks without additional penicillin. This patient has not been seen since June 1946, and the authors have no information as to the subsequent course. This case is considered a failure, due in all probability to too early operative interference in the post-acute stage of osteomyelitis with inadequate surgical removal of devitalized tissues.

Case 50–73. A man, twenty-six years old, had had hematogenous osteomyelitis of the left ilium at the age of ten. This was followed by many saucerizations and Orr dressings, maggots, and toxoid therapy. At the age of twenty the limb was disarticulated at the hip. The stump was revised on three occasions. Despite all the therapy, a draining sinus leading to the acetabulum persisted. This was complicated by severe amyloidosis, affecting primarily the kidneys. Examination showed multiple scars on the lateral aspect of the left ilium and a draining sinus more anteriorly. Roentgenograms showed several ossific shadows in the soft tissues opposite the acetabulum. On January 2, 1945, under antibiotic control, the scar over the lateral aspect of the stump was excised down to bone. Pus was encountered in the acetabulum. The soft-tissue scar was very dense and avascular, and only part of it and the overhanging rim of the acetabulum could be excised. A plastic procedure was carried out to make closure of the wound possible. The wound failed to heal and the sinus persisted. Several years later the patient died of uraemia. Failure was attributed to inadequate surgery, due to the extensiveness of the lesion and the severe amyloidosis affecting the kidneys.

Case 106–148, 106–159, 106–191. A woman, forty-eight years old, had had hematogenous osteomyelitis of the left femur for thirty years. Healing had occurred subsequent to operation, with recurrences nineteen, twelve, and seven years previously. Intervals of healing had followed the operative procedures, but persistent drainage had been present for the past seven years. Examination revealed a draining sinus on the lateral aspect and two healed scars on the medial aspect of the thigh. Roentgenograms showed extensive involvement of the lower three fourths of the femur. On December 11, 1945, a saucerization was performed under antibiotic control. Healing failed to occur, notwithstanding the administration of 3,720,000 units of penicillin. Repeated cultures revealed penicillin-resistant, coagulase-positive *Staphylococcus aureus* in addition to secondary invaders. Failure in this instance was attributed to inability to excise the soft-tissue scar, which was very extensive and avascular, and to a penicillin-resistant mixed infection.

On January 22, 1946, the saucerized area was revised under antibiotic control. An abscess was found in the lower portion of the previously saucerized area and in the posterior mass of soft tissues. Healing failed after the spontaneous evacuation of the hematoma, despite the administration of 2,940,000 units of penicillin. Cultures revealed penicillin-sensitive, coagulase-positive *Staphylococcus aureus* overgrown by secondary invaders. The failure was attributed to the density of the soft-tissue scar, which could not be excised, and the presence of secondary contaminants.

The patient was readmitted on June 5, 1946, with an acute exacerbation of the chronic lesion, which had been draining since the previous operation. Cultures of pus from the sinus disclosed the presence of secondary invading organisms. On June 7, the area was again saucerized under antibiotic control. A large abscess was found in the soft-tissue scar, in addition to many suspicious areas in the bone, which was extensively diseased. The soft-tissue scar could not be removed entirely. The wound was closed without drainage, and for the third time it failed to heal. Cultures revealed penicillin-resistant, coagulase-positive *Staphylococcus aureus* and secondary organisms. Failure was again attributed to the extensiveness of the scar and the inability to remove it.

CASE 125–179. A woman, fifty years old, gave a history of a hematogenous osteomyelitic lesion of a femur of twenty years' duration; there had been several operations and persistant drainage. Examination revealed extensive involvement of the lower half of the femur with a foul-smelling wound on the anterior aspect and draining sinus on the medial and lateral aspects. The knee was ankylosed. Wound cultures revealed a mixture of secondary invaders.

On March 12, 1946, the diseased area was saucerized, a skin plastic operation to permit closure of the wound was carried out, and the sinus were curetted. All incisions were closed under antibiotic control. The wound broke down because of necrosis of skin flaps, so that the underlying hematoma was exposed. Failure was attributed to inadequate skin closure, predominance of secondary invading micro-organisms resistant to penicillin, and excessive involvement of the osteomyelitic focus, making adequate surgery impossible. Failure had been anticipated and amputation had been primarily advised in the event that this conservative surgery did not succeed.

CASE 161–234. A man, forty-five years old, had an acute lesion of his left thigh for which seven doses of roentgenotherapy (1,250 r) had been administered elsewhere. Examination four weeks later by another surgeon revealed a fusiform swelling of the thigh with localized tenderness, local heat, and roentgenographic evidence of an osteomyelitic lesion. The temperature rose as high as 102 degrees. On September 7, 1946, the lesion was drained and saucerized, and the wound was packed with vaseline gauze. Penicillin was administered, a concentration of 1,000 units per cubic centimeter of saline locally and 50,000 units every three hours intramuscularly. Coagulase-positive *Staphylococcus aureus* was isolated from the culture taken at the time of operative intervention. A culture taken ten days later revealed coagulase-positive *Staphylococcus aureus* resistant to 25 units of penicillin, in addition to secondary contaminants. Large doses of various sulfonamide drugs and of streptomycin were added to the therapeutic regimen. Fluctuating temperatures persisted and a pathological fracture occurred during the fifth postoperative week.

When the patient was seen by the authors, on November 18, 1946, examination revealed an acutely ill man, with a temperature fluctuating to 102 degrees. Copious amounts of pus were draining from a wound, ten inches long and three inches wide, and a pathological fracture of the femur was in the depth of the wound. A culture grew coagulase-positive *Staphylococcus aureus* which was resistant to 50 units of penicillin, 12.5 micrograms of streptomycin, and 500 milligrams of sulfathiazole; *Bacillus pyocyaneus*, resistant to 100 micrograms of streptomycin; and secondary contaminants.

On December 3, the area was saucerized; the fracture ends were brought into alignment and impaction, resulting in considerable shortening of the femur. The wound was closed without drainage and the limb was immobilized in a plaster-of-Paris spica. Antibiotic control consisted of the intramuscular administration of 50,000 units of penicillin and 0.5 gram of streptomycin every three hours. One thousand units of penicillin and 1,000 micrograms of streptomycin per cubic centimeter of saline were used for topical application at the time of the operative procedure. The general condition of the patient improved greatly, and one month later the wound was healed except for a granulating area measuring one by one and one-half inches.

Three months later the patient was readmitted for change of plaster. Examination disclosed non-union of the femur and two small wounds about the knee. A small amount of drainage persisted for some time thereafter.

Failure to obtain healing by primary intention in this instance was attributed to the presence of an extensively mixed infection by organisms resistant to penicillin, streptomycin, and sulfonamides. The deep roentgenotherapy had undoubtedly interfered with the healing potentialities of the area involved. Notwithstanding the failure to obtain primary healing, the antibiotic therapy without a doubt modified the course of the disease for the better and averted what seemed to be an unavoidable amputation.

CASE 229–327. In a woman, twenty-nine years old, hematogenous osteomyelitis of the right tibia

had developed in September 1945, following an operation for removal of a fibroid. Despite a saucerization elsewhere and the administration of penicillin, drainage persisted. On April 27, 1948, the tibia was saucerized and the wound was closed without drainage under antibiotic control. The outcome was a failure, attributable to a mixed infection from organisms which developed a rapidly increasing resistance from 25 to 250 units of penicillin and from 25 to 625 micrograms of streptomycin.

CASE 239–339. A man, forty-seven years old, presented a hematogenous osteomyelitic lesion of nine years' duration, involving extensively the posterior portion of the left ilium and the adjacent area of the sacrum. Despite several operations and the administration of penicillin, the focus repeatedly broke down and drained pus. On January 10, 1948, the diseased portion of the ilium, which was greatly thickened, was resected; a soft-tissue sinus tract was found to extend into the interior of the pelvis. The entire tract could not be removed. The wound was closed without drainage and the antibiotic regimen was carried out. The outcome was a failure, due to the inability to institute adequate surgery and to the evacuation of the hematoma. This was complicated by the presence of a mixed infection.

CASE 244–346. A man, seventy-six years old, had had a compound fracture of the right foot sixty-two years previously, with healing following a surgical procedure. There was no further history of disability until six weeks prior to admission, when a sinus developed in the sole of the foot. Examination revealed a draining sinus, surrounded by a dense scar on the plantar aspect of the foot. Roentgenographic examination showed an extensive deformity of the mid-tarsus, which had fused into one bony mass with a large sequestrum in the soft tissues of the plantar aspect. A culture grew coagulase-positive *Staphylococcus aureus*, overgrown by *Bacillus proteus*.

On May 10, 1949, the sequestrum was removed under antibiotic control, and the wound was closed without drainage. No attempt was made to remove the soft-tissue scar. Necrosis of the wound margins was found at removal of the sutures, accompanied by maceration of the surrounding area. The patient was released one month later with the operative wound not entirely healed. This failure was attributed to inadequate removal of the soft-tissue scar and embarrassed circulation of the soft tissues involved.

CASE 248–350. A twenty-year-old woman sustained compound fractures of both tibiae and fibulae in April 1946 as a result of an automobile accident. In May, the right leg was amputated below the knee. At the first change of plaster on the left side, foot-drop was noted as well as a slough in the popliteal space. The wound over the tibia failed to heal and, despite three saucerizations, three sinus persisted.

Examination revealed an atrophic limb with multiple scars, paralytic equinovarus adduction deformity of the foot, ankylosis of the ankle joint in malposition, a half-inch ulcer over the lateral malleolus, three draining sinus along the anteromedial surface of the tibia with devitalized bone exposed in one opening, and a healed transverse scar in the popliteal space. The limb was somewhat dusky and oedematous. Roentgenograms showed malunited fractures of the tibia and fibula with overriding and angulation of the fragments and extensive osteomyelitic changes in the tibia. Cultures produced a mixed infection, resistant to both penicillin and streptomycin.

On October 12, 1949, under antibiotic control, the tibia was saucerized; all sinus tracts, one of which led through the posterior cortex into the popliteal space, were excised; and a skin plastic operation with releasing incisions was performed. All wounds save that resulting from the releasing incision over the lateral aspect of the leg were closed without drainage. A saline solution, containing 500 units of penicillin and 1,000 micrograms of streptomycin per cubic centimeter, was used during the operative procedure. The intramuscular injections consisted of 50,000 units of penicillin and 0.15 gram of streptomycin. The wound healed *per primam* except for a punctate area which drained persistently.

Despite this drainage, a double-wedge resection was performed six weeks later under antibiotic control for correction of the deformity of the foot. The wound healed *per primam* and bony union occurred. Recent examination revealed satisfactory reformation of the tibia, save for the presence of a sequestrum which should be removed in the near future. This over-all result, although eminently satisfactory to the patient, was considered a failure due to the persistence of a sinus and a sequestrum. The failure was attributed to the presence of a mixture of organisms, resistant to 25 units of penicillin and 625 micrograms of streptomycin, and to an incomplete surgical procedure.

A summation of all the lesions (Table II) shows that healing was induced in a total of eighty-four or 90 per cent. In only 3 (3 per cent.) was there any recurrence during the period of observation. One of these lesions was resaucerized with resultant healing by primary intention. Another patient was lost to follow-up, and the third is still under observation.

A consideration of percentile results on the basis of the number of saucerization procedures carried out (Table III) shows that, in seventy-eight of 100 saucerization opera-

TABLE II

HEALING OF CHRONIC OSTEOMYELITIC LESIONS IN EIGHTY-THREE PATIENTS SUBSEQUENT TO SAUCERIZATION, PRIMARY CLOSURE, AND ANTIBIOTIC CONTROL

Healing	No. of Lesions	Per cent.
By primary or delayed primary intention	73	79
After prolonged drainage	1	1
After additional procedure (pinch graft or dermoplasty)	5	5
By primary intention subsequent to resaucerization	5	5
Total lesions healed	84	90
Recurrences	3	3
Failed to heal after one or more saucerizations	9	10
Total	93	100

tions, healing was by primary or delayed primary intention; in 1 per cent. healing occurred after a prolonged period of drainage; and in 5 per cent. healing took place after additional grafting procedures. In 16 per cent. the lesions failed to heal.

It is evident, therefore, that the treatment of chronic osteomyelitis by thorough saucerization and primary wound closure without drainage, under antibiotic control (penicillin, streptomycin, or a combination of both), is the most effective method available at the present time. Eighty per cent. of surgically approachable lesions healed by primary or delayed primary intention; an additional 5 per cent. healed subsequent to additional skin-grafting procedures; and a further 5 per cent. healed subsequent to resaucerization of the lesion. In only 3 per cent. of these cases has there been recurrence over a period of observation varying up to five and one-half years. The 10 per cent. of failures are chiefly due to the presence of mixed infections, resistant micro-organisms, or deficient surgery, in some instances due to the nature of the lesion and in some instances to the inadequacy of the surgeon. Other disadvantages of the present use of penicillin and streptomycin are the necessity for frequent intramuscular injections; the presence and the development in certain instances of allergic and toxic manifestations to a degree which prohibits the use of these drugs; and, finally, the presence or the development of resistance on the part of the organisms to a degree which in some instances may make the antibiotic ineffective. It is because of these latter difficulties, even though they are of a relatively minor nature, that the authors are studying the efficacy of aureomycin* in the treatment of chronic osteomyelitis.

The lack of toxic and allergic manifestations incidental to the use of aureomycin and the ease with which it is administered orally or intravenously are well known. Furthermore, it has been shown by several investigators [22, 23] that the range of effectiveness of this antibiotic is much greater than that of penicillin or streptomycin, or of both together, thus making the drug more effective in the presence of mixed infections. It has been shown [23] that coagulase-positive *Staphylococcus aureus* is particularly sensitive to aureomycin, even though many of these organisms are resistant to both penicillin and streptomycin.

In tests of sensitivity to penicillin, aureomycin, and streptomycin, done in the bacteriology laboratory of the Hospital for Joint Diseases on forty-three strains of coagulase-positive *Staphylococcus aureus* isolated from twenty-seven patients with osteomyelitis, it was found that no parallel relationship existed between the degrees of susceptibility of the Staphylococci to the three antibiotics. Essentially all cultures of *Staphylococcus aureus* tested have been inhibited by aureomycin in a concentration of 1 micro-

* Aureomycin for oral and intravenous use was made available through the generosity of the Lederle Laboratories Division, American Cyanamid Company.

TABLE III

RESULTS OF 100 SAUCERIZATION PROCEDURES WITH PRIMARY CLOSURE

Results	Per cent.
Healing by primary or delayed primary intention	78
Healing after prolonged period of drainage	1
Healing after pinch graft or dermoplasty	5
Failed to heal	16
Total	100

gram or less per cubic centimeter; the majority of the strains were inhibited by 0.39 microgram or less.

Approximately 70 per cent. of strains of *Staphylococcus aureus* which acquired resistance to penicillin during treatment have retained their resistance to this antibiotic for periods of four to six years. Since strains which acquired resistance to penicillin *in vivo* remained virulent, they pose the theoretical problem of the value of further treatment with penicillin in those instances where resistance to this antibiotic has developed during therapy. These strains, as well as those strains which are naturally resistant to penicillin, are sensitive to aureomycin.

Using the techniques described by A. C. Dornbush, the authors have been able to confirm the fact reported by Dowling and his associates [20, 21] that blood levels ranging up to 4 micrograms per cubic centimeter can be produced and maintained in adults by the oral administration of 250 to 500 milligrams of aureomycin. In view of these findings, we have modified our management of chronic osteomyelitic lesions to allow for the oral administration of 500 milligrams of aureomycin every six hours for twenty-four hours before operative interference; an intravenous infusion of 1,000 cubic centimeters of saline containing 500 milligrams of the drug during the operative procedure; and 500 milligrams thereafter by mouth every six hours, depending upon the patient's tolerance of oral ingestion. We also apply the drug locally during closure of the wound in the approximate concentration of 0.5 milligram per cubic centimeter of saline. On several occasions this topical administration was also used in the unanaesthetized patient without any evident discomfort or irritation, despite the high pH level of the solution.

Our experience with aureomycin is only of recent date, and the cases treated with this antibiotic are too few in number to warrant a detailed analysis. Suffice it to say that our findings have been most encouraging, and in several instances aureomycin has converted what would have been unavoidable failure, under the penicillin and streptomycin regimens, to a successful outcome. The greater range of effectiveness of aureomycin, the lack of the development of resistance on the part of the offending organisms, the almost negligible development of allergic and toxic manifestations on the part of the patient, and the ease with which the drug is administered give promise of further improvement in the therapy of chronic osteomyelitis.

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DISCUSSION

Dr. Guy A. Caldwell, New Orleans, Louisiana: Dr. Buchman's report supplements our knowledge of the management of chronic osteomyelitis. He has given the results obtained in eighty-three patients with ninety-three lesions treated by well recognized surgical procedures, combined with the administration of antibiotics, including aureomycin. The selection of cases, preoperative preparation, operative technique, and postoperative treatment are based on sound surgical principles, and his results compare favorably with those of others who have followed the same general plan.

In 1943 and 1944, we carried out a similar project at Tulane University and Charity Hospital in New Orleans for the National Research Council and, in 1945, reported results in forty-three cases of osteomyelitis of the long bones. There were twenty-one cases of localized osteomyelitis, of which 80.4 per cent. healed *per primam;* however, in only 61.8 per cent. did the lesions remain healed after six to twelve months. There were twenty-two cases of hematogenous osteomyelitis, including nine tibiae, three femora, and five humeri. Of these, 81 per cent. had primary healing, but in only 63 per cent. did the lesions remain perfectly healed after five to eight months. Analysis of failures to heal in these groups indicated that incomplete removal of cicatrized or infected bone and skin coverage under too great tension were the important causes. Bacterial complication from mixed infection or penicillin-resistant organisms was considered to be a factor in 15 per cent. of the cases.

Since 1945, many additional patients have been treated by the same method with similar results. Somewhat better results were obtained in the later series, probably because our operative technique had improved and we had sufficient penicillin to give adequate doses over a period of several weeks. However, some lesions still heal primarily, but recur after a period of months or years.

Some of the most difficult cases have been those with large cavities in the lower femur or tibia, in which

it is not possible to saucerize the bone or flatten the soft parts and skin against the raw bone. In several such cases I have transplanted enough iliac bone to fill the cavity and have secured healing after one or more previous failures. The radical saucerization of two chronic wounds with involvement of the shafts of femora resulted in pathological fractures through the remaining brittle bone, followed by non-union and sinus; in both, amputation was subsequently required.

Probably there will always be some patients with chronic osteomyelitis, but the number should be small because of the present tendency to administer penicillin promptly in all cases of suspected osteomyelitis. Early and adequate treatment of the acute disease, even when there is extensive involvement of bone, will prevent chronic osteomyelitis in the great majority of cases. Therefore, we may expect few chronic cases after a few more years, and probably most of these will be of the localized exogenous type.

DR. J. ALBERT KEY, ST. LOUIS, MISSOURI: This is a very good paper and the results are about what most of us are now getting in these cases.

I am very much interested in the authors' views on organization of the hematoma. Apparently they like hematomata. I still try to avoid them. The less dead space one leaves, the more apt the wound is to heal.

Some years ago, I tried filling the dead space with oxidized cellulose. That doesn't work in the presence of infection, nor do grafts from living bones. In my experience nothing is more likely to become infected than a bone graft, and this is especially true of cancellous bone. We have had better success with merthiolate bone from our bone bank.

Some of these cavities are not amenable to saucerization and cannot be sterilized. I am treating one now,—a big bone cyst. We filled it with rib and chip grafts; it became infected. I cleaned it out as well as I could, sewed it up according to Dr. Buchman's principles, as we all do, and it healed. In spite of heroic treatment with antibiotics, a sinus developed in a few weeks.

Recently I used the procedure of Dr. Bennett and Dr. Johnson for the treatment of giant-cell tumors. The femur is cut across just above the condyles and the proximal fragment is pushed down into the cavity from which the tumor has been removed, thus shortening the limb about two inches and eliminating the dead space.

There will still be cases in which amputation is the wise procedure. The bacteria can be inhibited by antibiotics, but grow again when the antibiotics are stopped.

DR. LOUIS G. HOWARD, BOSTON, MASSACHUSETTS: Our work in Boston closely parallels Dr. Buchman's; we published the results on those patients operated upon up to March 1947.

I doubt if local administration of the antibiotics has very much effect. The valuable effect of antibiotic therapy in osteomyelitis is the capillary-bed supply at the base of the wound, resulting from intramuscular or oral administration.

It is our habit to clean out thoroughly all diseased tissue, including sinus tracts, unhealthy bone, sequestra, and granulation tissue, at the same time preserving as much healthy bone as possible.

We have also treated six cases without local administration of antibiotic therapy, and the patients have done just as well as those who received it locally. That is experimental, and, until we know more about it, we should not teach it.

DR. FRANK D. DICKSON, KANSAS CITY, MISSOURI: To me, the surgery is the important thing. It must include not only removal of all necrotic and infected bone, but, unless all scar tissue in the soft parts is completely resected at the same time, the results will not be what they should. The only substance with which to fill a bone cavity so as to avoid hematoma is living muscle. When we do saucerization, we never stop until we have a normal, healthy muscle to put into that cavity. Surrounding any bone infection there is a sclerotic zone which has a poor blood supply, and, unless that blood supply is augmented, there will be quite a few failures.

Perhaps there is a little too much pessimism about the ability to saucerize cavities. I believe that we can be much more ruthless than some of us think.

I am going to bring down a good deal of criticism on my head by saying this: When we started our work, ten years ago, we powdered the wound with sulfathiazole, which in those days was the most effective antibiotic against the Streptococcus, and closed it. My own impression is that our results are a little better than those reported, so that we continue today to use sulfathiazole—not penicillin, aureomycin, or anything else—and to put on a good snug-fitting cast. We are getting immediate healing in a high percentage of cases.

DR. BUCHMAN (closing): I want to thank all of the discussors for their kind remarks.

There appears to be general agreement as to the effectiveness of primary closure of the saucerized wound in the treatment of chronic osteomyelitis. Several variations of technique have been presented. I cannot discuss all of these in the few minutes available; nevertheless I want to emphasize that, with the use of the

(*Continued on page 130*)

TUBERCULOSIS OF THE SACRO-ILIAC JOINT

A Review of Seventy-five Cases

BY STANLEY T. SOHOLT, M.D., MIDDLEBORO, MASSACHUSETTS

*From the Lakeville State Sanatorium *, Middleboro*

Tuberculosis of the sacro-iliac joint is not ordinarily regarded as a rare disease, but few reports embodying a sizable series of cases are to be found in the literature. Hence, it is fair to assume that the experience of any one individual is limited. The purpose of this paper is to present data from a review of seventy-five cases of this disease, in the hope that this information may add to that already gained from the few previous analyses. The most thorough and largest of these is that of Seddon and Strange, who, in 1940, presented 176 cases. Petter, in 1934, reviewed thirty-one cases; Thompson, in 1938, twenty-three cases; and Pollack and Bosworth, in 1942, forty-four cases. Other authors have made mention of this disease in case reviews pertaining to bone and joint tuberculosis.

Separate consideration of the sacro-iliac joint is particularly indicated in any study of bone and joint tuberculosis, because of the unique anatomy and function of the joint,

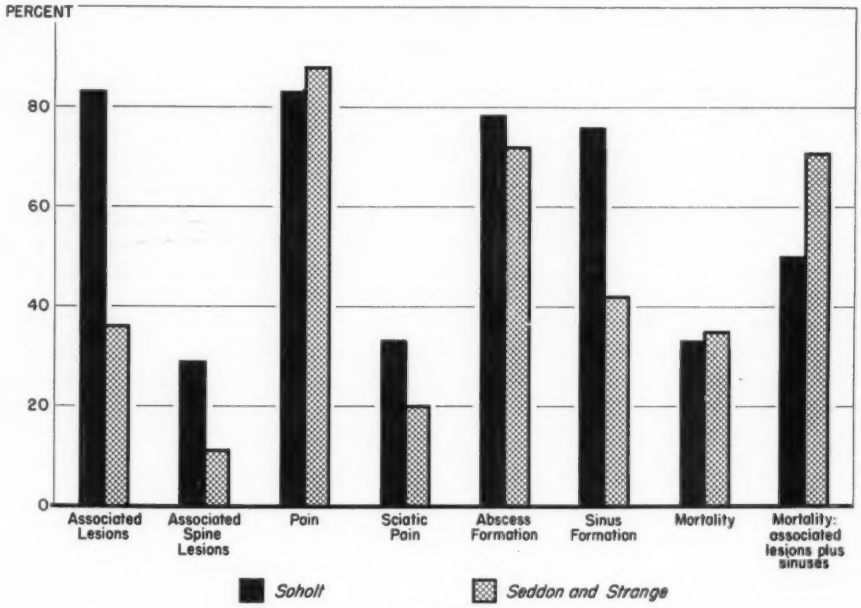

CHART I

A comparison of the findings of the present study with those of Seddon and Strange.

and because the diagnosis of disease located therein is frequently so delayed that definitive treatment is instituted late in the course of the disease. The joint is amphiarthrodial with two articular surfaces of hyaline cartilage, united by patches of fibrocartilage. Its primary function with its many strong ligaments is to provide a stable means of weight transmission from the spine. It also allows a very slight range of motion.

* Service of George W. Van Gorder, M.D.

TABLE I

ASSOCIATED TUBERCULOUS LESIONS *

Lesions	No. of Patients**	Per cent.
Bone and joint	35	47
Pulmonary	33	44
Lymph gland	11	15
Peritoneal	9	12
Renal	6	8
Meningeal	4	5

* Present in sixty-two of seventy-five cases.

** Patients with more than one type of lesion are included in more than one group.

The material for this paper is derived from a study of seventy-five cases of tuberculosis of the sacro-iliac joint, seen at the Lakeville State Sanatorium during the twenty-three years from 1925 to 1948. In this group each case had what was regarded as roentgenographic evidence of sacro-iliac tuberculosis, and sixty had positive bacteriological and pathological proof of the disease, alone or in combination.

In presenting this material, an attempt is made wherever possible to compare the findings in this series with those of Seddon and Strange (Chart I).

MOST IMPORTANT FEATURES OF THE DISEASE

Incidence

An important feature of this disease is that it occurs in young adults and is thereby distinguished from all other forms of joint tuberculosis. Fifty-nine patients (79 per cent.) were between twenty and forty years old. The youngest was seven years of age and the oldest fifty-seven, the average being thirty years. (Seddon and Strange found that 80 per cent. of the patients in their series were between sixteen and thirty-five years of age, the average being about twenty-five years.) The race and sex distribution was not remarkable: sixty-six white patients, eight Negroes, and one Chinese; forty-one (55 per cent.) were females. The disease was bilateral in seven cases.

Associated Lesions

Tuberculous lesions elsewhere in the body were commonly associated with sacro-iliac disease (Table I). Other authors [1,6,7,8,10] have reported the presence of associated

TABLE II

ASSOCIATED BONE AND JOINT LESIONS IN THIRTY-FIVE CASES

Site	No. of Patients *	Per cent.
Spine	22	29
Hip	10	13
Ankle	3	4
Pubis	2	
Wrist	2	
Trochanter	1	
Radius	1	
Humerus	1	
Rib	1	
Zygoma	1	
Fibula	1	

* Patients with more than one osseous lesion are included in more than one group.

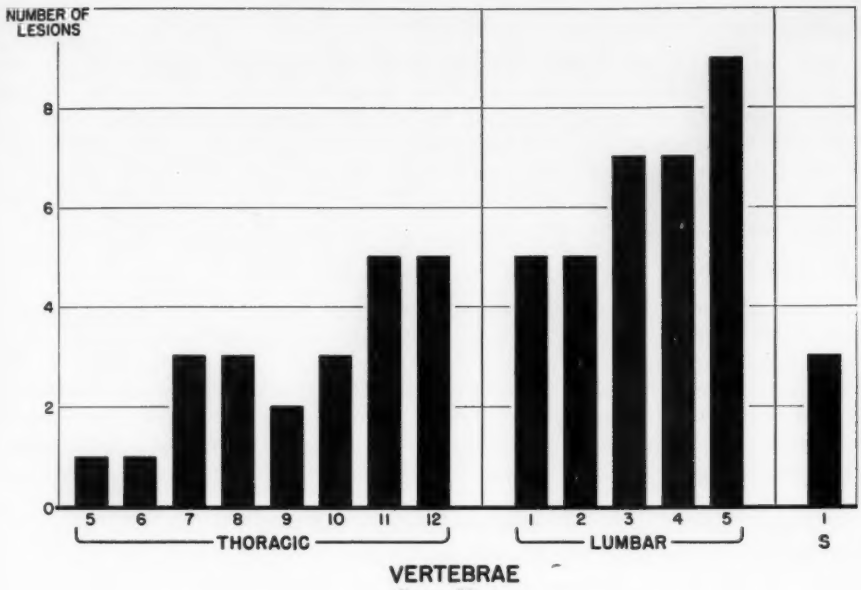

CHART II

Incidence of associated vertebral tuberculosis according to level.

lesions in from 36 per cent. to 84 per cent. of their cases. In this series of seventy-five patients, sixty-two (83 per cent.) had tuberculosis elsewhere in the body. In twenty-seven cases (36 per cent.) there was a single associated tuberculous condition; in thirty-five (47 per cent.), two or more.

Associated skeletal lesions were found in thirty-five cases or 47 per cent. (Table II). The most common site was the spine, which was involved in twenty-two cases (29 per cent.),—nearly three times as often as has been described elsewhere [8]. Next in frequency came the hip, in ten cases (13 per cent.). It has been suggested that the psoas muscle serves as a channel for the spread of the infection between the lumbar spine, the sacro-iliac joint, and the hip. The present study confirms the opinion, previously expressed by others, that in sacro-iliac tuberculosis there is a decided increase in the incidence of concomitant involvement of the lower lumbar vertebrae. The distribution of the lesions in individual vertebrae is indicated in Chart II; the fifth lumbar vertebra is most commonly implicated.

Onset

The onset of the disease was usually characterized by the appearance of pain in the posterior iliac or hip region. Pain was the first symptom in sixty-two cases (83 per cent.). It is important that sciatic radiation was present in twenty-five cases (33 per cent.), since this symptom has become popularly associated with lesions of the lumbosacral region. Four of these patients had associated lumbar lesions which might have caused the sciatic pain; however, in the remaining twenty-one cases there was no involvement of the lumbar spine. Swelling and abscess formation without pain occurred as the initial symptom in six cases (8 per cent.). In the remaining seven cases the onset of the disease was insidious without pain or swelling, and the diagnosis was made by routine roentgenographic examination. A history of trauma was occasionally elicited, but generally the relation of trauma to the onset of symptoms was vague and unconvincing. The first symptom occurred during pregnancy in three cases, and immediately after delivery in one.

Abscess Formation

The formation of an abscess during the course of the disease occurred in fifty-nine (79 per cent.) of the seventy-five cases. Most often the abscess was directly over the sacro-iliac joint; the second most common site was the inguinal region. The abscess subsided without drainage in only two instances; in the remaining fifty-seven patients (76 per cent.) draining sinus developed,—forty-six spontaneously and eleven postoperatively. Seddon and Strange reported abscess formation without sinus in 30 per cent., and sinus formation in only 42 per cent.

Pathological Proof

By examination of material from the diseased sacro-iliac joint itself, either bacteriological or pathological proof (or both) of the presence of tuberculosis was obtained in forty-two cases (56 per cent.). Similar proof of tuberculosis elsewhere in the body was obtained in eighteen additional cases (24 per cent.). Thus, a total of sixty patients (80 per cent.) in this series of seventy-five cases had bacteriological and pathological proof of tuberculosis, either alone or in combination (only ten had pathological proof alone). Seddon and Strange reported "complete pathological proof" in 28 per cent. of their cases.

ROENTGENOGRAPHIC FINDINGS

The presumptive diagnosis of sacro-iliac tuberculosis is made primarily by the appearance of certain roentgenographic changes. This is usually confirmed by bacteriological and pathological studies. Demonstrable roentgenographic alterations in the joint were present in all cases in this series.

Early Changes: The earliest change consists of haziness or loss of definition of the joint line (Fig. 2-A). There then appears irregularity of the articular surfaces with areas of erosion (Fig. 1-A). If the disease subsides at this point, the joint space becomes narrowed and the adjacent bone is more dense. However, if the infection progresses, bone destruction becomes more marked and cavitation may develop.

Later Changes: In thirty-two instances, roentgenograms were available for examination after three or more years. All of these films had been interpreted by the hospital roentgenologist, and were reviewed by the author. The average length of roentgenographic follow-up was eight and one-half years. Of the thirty-two cases, thirty (94 per cent.) showed bony ankylosis of the involved sacro-iliac joints. The two remaining cases showed increased density about the joint without osseous fusion. Four of the patients in whom ankylosis of the sacro-iliac joint developed, and one in whom it did not develop, died subsequently. Seven patients who were followed only from one to three years also had ankylosis, as shown by roentgenogram.

PROGNOSIS

The findings of the present study show that this disease has a poor prognosis, a fact which has also been demonstrated in previous reports [1,8,10]. The mortality has been high and hospitalization has been lengthy. In this series, twenty-five patients died in the Hospital, a mortality of 33 per cent.

Causes of Death

An analysis revealed that seventeen (68 per cent.) of the twenty-five patients who died in the Hospital succumbed to overwhelming infection, amyloidosis, and inanition due to long-standing infection; four died of tuberculous meningitis; three of severe pulmonary tuberculosis; and one of uraemia. Six additional patients are known to have died since discharge,—three of tuberculosis, one of an unrelated condition, and two of unknown causes.

TABLE III

CHRONOLOGICAL DISTRIBUTION OF MORTALITY

Time of Death	No. of Patients	No. of Deaths	Mortality (*Per cent.*)
1926 to 1930	23	15	65
1931 to 1935	21	5	24
1936 to 1940	12	4	33
1941 to 1948	19	1	5
Total	75	25	33

Age

Seddon and Strange found the highest mortality rate in the older age groups. In the present series this did not appear to be true, since the age distribution of deaths very closely paralleled the age incidence of the disease. The average age at the time of death was thirty-one years, the youngest patient being ten years and the oldest fifty-seven.

Time after Onset

The average length of time from the onset of symptoms until the patient died in the Hospital was thirty-eight months.

Chronological Distribution of Deaths

Investigation showed a marked decline in the mortality rate in recent years (Table III). Of forty-four patients admitted from 1926 to 1935, inclusive, twenty (45 per cent.) died in the Hospital. Of thirty-one patients admitted from 1936 to 1948, five (16 per cent.) died in the Hospital. Only one of these deaths occurred after 1940.

This declining mortality rate demonstrates that the total figures do not give a true picture of the present-day prognosis of sacro-iliac tuberculosis.

Sinus

It has been felt by most authors that the secondary infection which always accompanies a draining sinus has a serious effect on the prognosis. This belief is supported by the following observations:

Of the seventy-five patients in this series, eighteen (24 per cent.) did not have draining sinus during their stay in the Hospital; none of these patients died. Of the remaining

TABLE IV

THE EFFECT OF SINUS AND ASSOCIATED LESIONS ON MORTALITY

Type of Lesion	No. of Patients	No. of Deaths	Per cent.
Without sinus			
Isolated sacro-iliac disease	3	0	0
Other tuberculous lesions	15	0	0
With sinus			
Isolated sacro-iliac disease	10	3	30
Other tuberculous lesions	47	22	47
Total	75	25	33
With multiple sinus (included above)	14	7	50

TABLE V

SUMMARY OF FINDINGS OF TWENTY-NINE SURVIVORS FOLLOWED THREE YEARS OR MORE

Patient	Sex	Age	Length of Follow-up (*Years*)	Operative Fusion	Pain	Drainage	X-Ray Appearance	Comment
M. A.	F.	48	13	Yes	No	Yes	Bony ankylosis; sequestrum present	Works 8 hours daily
M. A.	F.	35	18	Yes	No	No	Not available	Works 4 hours daily
J. B.	M.	29	6	No	No	No	Bony ankylosis	Not working; feels well
N. B.	F.	50	13	Yes	No	No	Bony ankylosis	Works 7½ hours daily
C. B.	F.	29	18	No	No	No	Bony ankylosis	Works 8½ hours daily
C. B.	M.	55	15	No	Yes	No	Bony ankylosis	Works 5 hours daily
E. C.	M.	64	21	No	No	No	Not available	Works 8 hours daily
J. C.	F.	36	12	Yes	No	No	Bony ankylosis	Housework; has had 2 normal deliveries
L. D.	M.	34	4	Yes	No	No	Bony ankylosis	Recovering from fracture of tibia
V. E.	F.	45	19	No	No	No	Bony ankylosis	Works 8 hours daily
J. F.	M.	44	20	No	No	No	Bony ankylosis	Works 8 hours daily
K. G.	M.	39	4	Yes	No	No	Bony ankylosis	Not working
J. G.	F.	39	16	Yes	No	No	Bony ankylosis	Works only part time because of hernia
A. G.	F.	48	7	Yes	No	No	Bony ankylosis	Housework
F. J.	F.	28	4	Yes	No	No	Bony ankylosis	Not working (looking for work)
M. K.	F.	38	18	Yes	No	No	Bony ankylosis	Works 8 hours daily
A. LaC.	M.	35	16	Yes	No	No	Bony ankylosis	Works 9 hours daily
S. L.	F.	27	3	Yes	No	No	Bony ankylosis	Housework
S. M.	M.	57	18	Yes	No	No	Bony ankylosis	Works 8 hours daily
E. M.	F.	40	3	Yes	No	No	Bony ankylosis	Housework
P. P.	F.	36	8	Yes	No	No	Bony ankylosis	Works 7 hours daily
M. R.	F.	36	15	Yes	No	No	Bony ankylosis	Works 8 hours daily
D. R.	F.	38	18	Yes	No	No	Bony ankylosis	Housework; has had 3 normal deliveries
E. S.	F.	58	10	Yes	Yes	No	Bony ankylosis	Works 15 hours daily
E. S.	M.	44	16	No	No	Yes	Bony ankylosis	Works 7 hours daily
R. T.	M.	19	3	Yes	No	No	Bony ankylosis	Not working
L. V.	F.	36	6	No	No	No	Increased density; no ankylosis	Housework
M. W.	F.	24	5	No	Yes	No	Bony ankylosis	Works 8 hours daily
D. W.	F.	28	3	No	No	No	Bony ankylosis	Works 8 hours daily

fifty-seven patients who had sinus, twenty-five (44 per cent.) died (Table IV). Of all patients with draining sinus, fourteen had multiple sinus; seven of these (50 per cent.) died in the Hospital.

Thirty-two patients with sinus were discharged from the Hospital; twenty-three were followed for two or more years. Only three of them had persistent drainage; two were followed for thirteen and sixteen years, respectively, and were working full time.

Return to Work

In seeking further data relating to the prognosis in this disease, it is of importance to learn about a patient's ability to work, as well as his residual symptoms. A follow-up of three years or more (an average of eleven and one-half years) was obtained in twenty-nine cases, or 66 per cent. of the forty-four survivors (Table V). Twenty-four patients had no pain or drainage; three had mild pain, but no drainage; and two had persistent drainage, but no pain.

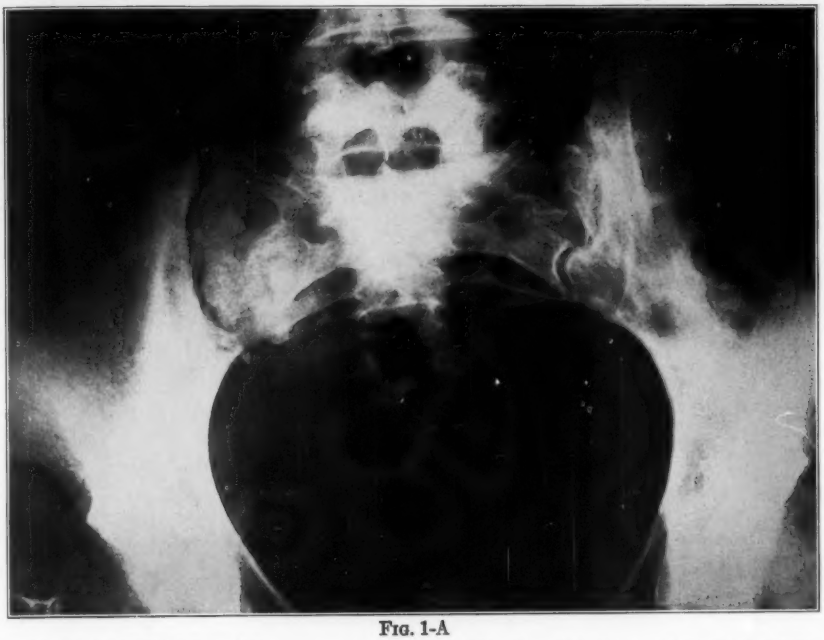

FIG. 1-A

March 27, 1943. F. J., a white woman, aged twenty-eight, showed haziness and irregular destruction of the surfaces of the right sacro-iliac joint.

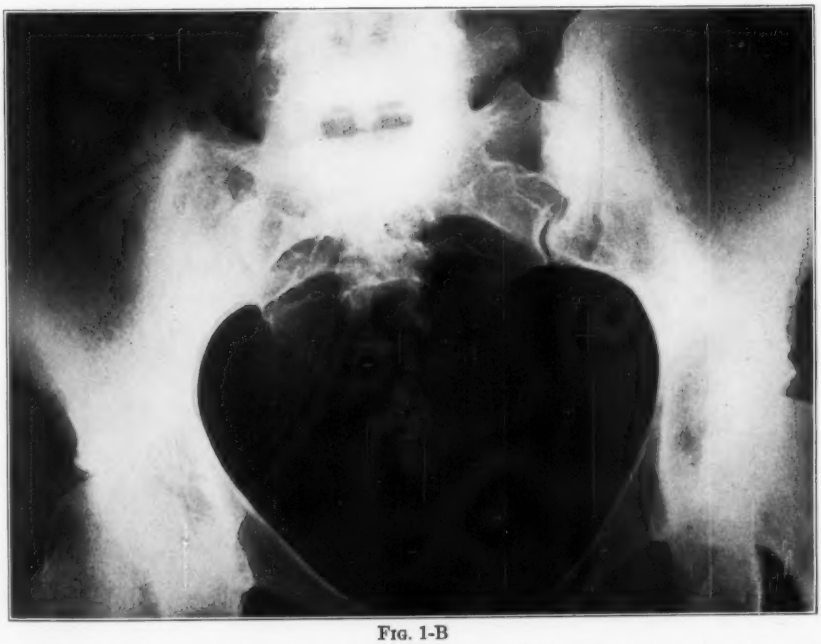

FIG. 1-B

April 1, 1949, five years after operative fusion. There was solid bony ankylosis of the right sacro-iliac joint. Patient was free of symptoms.

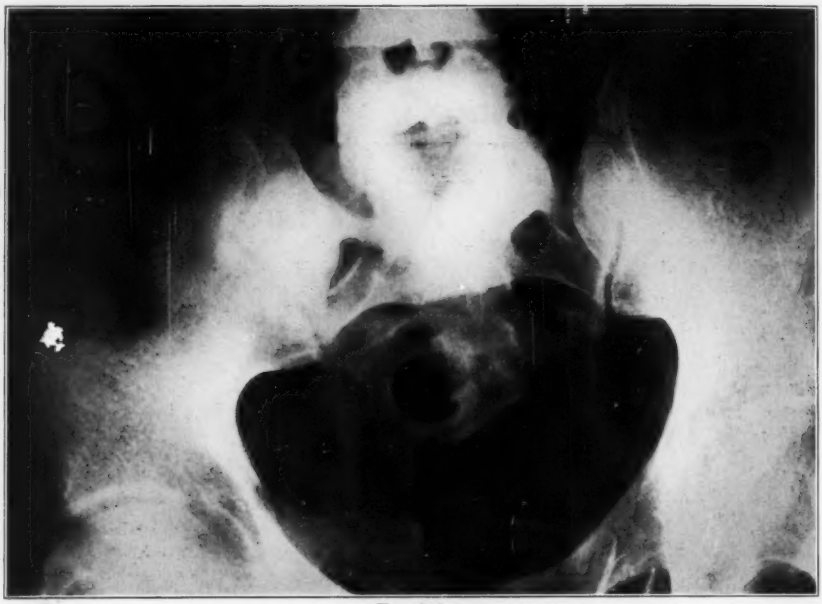

FIG. 2-A

March 3, 1930. J. F., a white male, aged forty-four, presented irregular destruction of the right sacro-iliac joint.

FIG. 2-B

April 1, 1949. There was solid bony ankylosis of the right sacro-iliac joint, without operation. Patient had no symptoms.

Twenty-four patients were working in wage-earning jobs or doing all their own housework. The remaining five patients were not working; however, one was looking for a job

FIG. 3-A

July 16, 1936. Roentgenogram of N. B., a white female, aged fifty, six months after operation. There was haziness of the upper half of the right sacro-iliac joint, with a bone graft across the lower half of the joint.

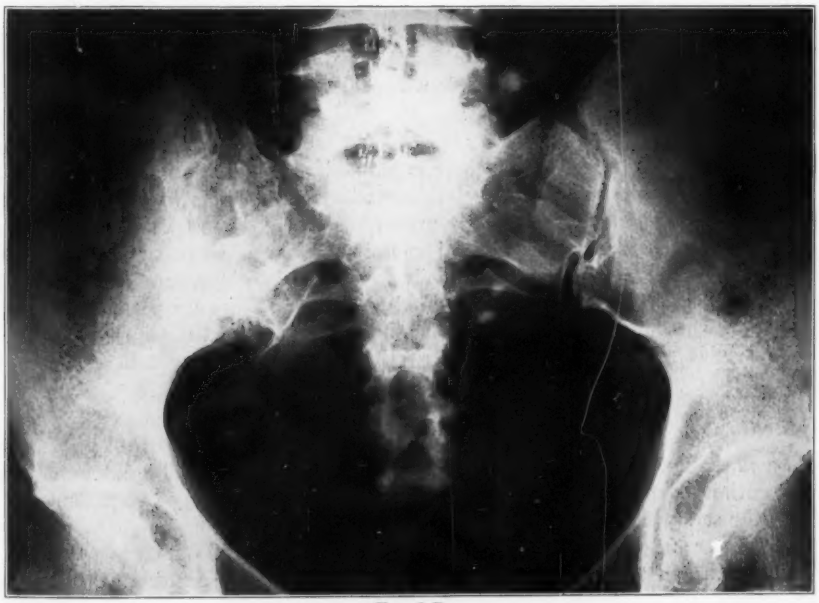

FIG. 3-B

March 22, 1949. There was bony ankylosis of the lower half of the right sacro-iliac joint with partial obliteration of the upper half. Patient was asymptomatic.

TABLE VI

FUSION OPERATION COMPARED WITH NON-OPERATIVE TREATMENT

Treatment	No. of Patients	No. of Deaths	Per cent.	Average Length of Hospitalization (*Months*)
Operative	32	6	19	24
Non-operative	43	19	44	28
Total	75	25	33	26

and another was recovering from a fracture of the tibia at the time the study was conducted.

Of seventeen patients followed for ten or more years, all were working, two had persistent drainage, and all had solid bony fusion as demonstrated by roentgenogram.

Bilateral Disease

The disease involved both sacro-iliac joints in seven cases. Two patients died in the Hospital; the three who had been followed more than three years are working and show bony fusion by roentgenogram.

Pregnancies

Two patients subsequently had normal pregnancies and deliveries, without apparent recurrence of the disease.

TREATMENT

Tuberculosis of the sacro-iliac joint must be treated as a manifestation of a systemic disease. Bed rest and good nutrition have long been recognized as essential in the proper management of tuberculosis. Adequate immobilization in plaster places the sacro-iliac joint at rest, but this may not be feasible when there are multiple draining sinus.

Antibiotics

Chemotherapy has recently become a valuable adjunct in the treatment of this condition. The antibiotics which combat secondary infection have made the draining sinus a much less ominous sign. Streptomycin has received most attention, because of its apparent direct action on the tuberculous lesion. Seven patients in this series received streptomycin,—0.5 gram once or twice daily for varying periods, ranging from seven to seventeen weeks. The results were encouraging, but the follow-up period has not been long enough. The value of streptomycin has been demonstrated in tuberculosis of other bones and joints; hence, at present its routine use in sacro-iliac tuberculosis can be accepted.

Operative Fusion

The efficacy of operative fusion in sacro-iliac tuberculosis is still a matter of debate, and the author's findings do little to help settle the question. Thirty-two patients (43 per cent.) in the series had operative fusion of the involved joints.

Eleven were fused by the Campbell method, ten by the Smith-Petersen method, and two by erasion and the placement of bone chips. Adequate description of the operative procedure was not available in the remaining eight cases. Six of the patients operated upon subsequently died in the Hospital, a mortality of 19 per cent. (Table VI). This is in contrast to a mortality of 44 per cent. among the forty-three patients not operated

upon. However, this can be accounted for, to some degree, by the selection of cases for operation. Twenty-three of the thirty-two patients operated upon had no draining sinus, seven had no associated tuberculosis elsewhere in the body, and only three had multiple sinus. Seddon and Strange also showed that careful selection of cases for operation could account for better end results.

Nineteen of the patients operated upon were followed for three or more years (an average of eleven years). Eighteen had solid bony fusion as shown by roentgenogram, although one of them had slight but persistent drainage at the end of ten years. The remaining patient was asymptomatic after eighteen years, but roentgenograms were not available.

Ten of the patients not operated upon were followed for a similar length of time. Eight had roentgenographic evidence of bony ankylosis, but one still had mild drainage after sixteen years. One patient had increased density about the involved joint, without fusion; the remaining patient had no symptoms after twenty-one years, but late roentgenograms were not available.

It has been felt that ankylosis of the sacro-iliac joint in this disease develops only after operation or with secondary infection caused by a draining sinus. In this series, however, bony ankylosis of the involved sacro-iliac joint developed in four patients who had no sacro-iliac operation or draining sinus. There was also one patient with bilateral disease, who had no draining sinus and had operative fusion on one side only; eleven years later she had bony ankylosis of both sacro-iliac joints.

These findings would tend to support the contention that the usual end result in patients surviving sacro-iliac tuberculosis, with or without operation, is bony ankylosis. This is the desired result, but some authors feel that this effect is more certainly and rapidly achieved by operative fusion in many cases.

CONCLUSIONS

Because this series consisted of only seventy-five cases, undue significance should not be attached to the facts presented. However, some of the data in this review, when added to those of other authors, may serve to depict certain features regarding the nature, treatment, and outcome of the disease.

The following characteristics of tuberculosis of the sacro-iliac joint have been demonstrated:

1. The disease occurred in young adults, usually between the ages of twenty and forty years (79 per cent.).
2. It was commonly associated with tuberculosis in other parts of the body (83 per cent.).
3. Pain was the most common initial symptom (83 per cent.).
4. Abscess formation occurred frequently (79 per cent.), usually followed by the formation of a draining sinus.
5. The usual roentgenographic appearance late in the disease was bony ankylosis (in 94 per cent. of cases followed three years or more).
6. The mortality was higher when there was a draining sinus (44 per cent.).
7. The data suggest, but do not prove, that operative fusion of the involved sacro-iliac joint is of value in hastening the desired end result.

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DISCUSSIONS

The Surgical Management of Chronic Osteomyelitis

(*Continued from page 118*)

technique I have described, I have seen only three recurrences in a group of ninety-three lesions over a period of five and one-half years.

With reference to the hematoma: The whole principle in the treatment of osteomyelitis, according to this method, is to depend for healing upon a hematoma filling the cavity. The implantation of muscle into a cavity isn't always feasible, nor is it desirable. Many saucerized wounds are sufficiently large and their location is such that there are no muscles available for transplantation into the cavity. Furthermore, when a muscle is transplanted into a cavity, it loses its function and becomes transformed into a mass of scar tissue which may be a focus of infection. The use of bone implants is unwarranted, for it is difficult enough to keep these wounds sterile and to obtain healing by primary intention without the introduction of foreign bodies. Furthermore, there is almost always sufficient bone formation during the course of healing to make the introduction of bone grafts unnecessary.

In the early phase of this work, I used gelfoam and oxidized gauze in an effort to fill these cavities. The wounds did not heal as well under these conditions. The best results were found in those instances in which healing took place by means of the hematoma.

I used plaster-of-Paris support in only one of these cases. The purpose of the compression bandage is to give support to the circulation of the limb, and, since the success of treatment in chronic osteomyelitis depends upon satisfactory revascularization of the area, the application of a compression bandage is most effective.

During the operative procedure, I excise as much of the diseased soft-tissue scar and bone as possible; nevertheless I have not had a single instance of fracture of the bone as the result of failure to use plaster-of-Paris support. The omission of plaster-of-Paris is desirable, because it avoids immobilization of the adjacent joints and makes possible an excellent range of motion.

Local application of the antibiotics is used only at the time of the operation. Subsequently, even if there is drainage, antibiotics are not injected into the sinus tract. The wound is allowed to heal without drainage. After the hematoma has been drained, a cavity develops, and healing thereafter can only occur by secondary intention.

The use of the sulfonamides was advocated prior to introduction of the antibiotics. They are still being advocated by some observers. Bacteriological studies have shown that the Staphylococcus is usually resistant to the sulfonamides, and that those strains which are sensitive readily develop resistance during therapy. These observations show that the sulfonamide drugs are generally ineffective.

TUBERCULOSIS OF THE HIP

A Follow-up Study of Fifty-eight Cases with Special Reference to Fusion Results in Young Children

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Fifty-eight unselected cases of tuberculosis of the hip, treated at the New York Orthopaedic Dispensary and Hospital between 1932 and 1946, formed the basis of this study, both from clinical and roentgenographic points of view. The cases are divided into two main age groups: Of the thirty-five children, ranging from one year and nine months to fourteen years of age, thirty were five years of age, or less, when first seen in this Clinic; and of the twenty-three adults from twenty-one to fifty years of age, thirteen became diseased in early childhood and ten in adult life; one of these had a bilateral hip involvement, with one hip affected in childhood and the other in adult life. In other words, among the adults, fourteen hips were diseased in early childhood, while ten became diseased in adult life. There are no cases in this series between the ages of fifteen and twenty.

Of the thirty-five patients in the children's group, thirty-three were treated by fusion operations, one was transferred to another hospital after exploratory arthrotomy, while in the other a spontaneous fusion resulted. In the group of twenty-three adults, with twenty-four hips involved, twenty-three hips were operated upon to obtain fusion and the remaining hip had had a spontaneous fusion when first seen in the Clinic.

The diagnosis was established by both tissue section and guinea-pig inoculation in thirty-six cases, by tissue section alone in nine, by guinea pig alone in five, and was not confirmed in eight cases. Many of the proved cases also had positive cultures for tubercle bacilli. In the eight unproved cases, the clinical and roentgenographic findings were such as to make an error in the diagnosis extremely unlikely. Other cases, seen during this period, in which there was a presumptive diagnosis of tuberculosis, but in which the

TABLE I

Deaths in Series

No.	Name	Age (*Years*)	Onset	Operations	Time of Death after Last Operation	Result of Fusion	Cause
1.	W. J.	4	12 months	1 exploratory 2 fusions	42½ months	Solid	Tuberculous meningitis
2.	L. H.	8	7 months	1 fusion	37 months	No fusion	Miliary tuberculosis and amyloidosis
3.	S. T.	1¾	6 months	3 fusions	22 months	Beginning fusion	Unknown
4.	McC. F.	28	5 years	1 fusion 1 sequestrectomy	7½ months	Beginning fusion	Unknown
5.	F. M.	29	2½ years	1 spine fusion 1 hip fusion	1½ months	No fusion	Miliary tuberculosis
6.	S. A.	32	2 years	1 fusion 1 exploratory	18 days	No fusion	Streptococcal septicaemia and active hip tuberculosis

TABLE II

DISTRIBUTION OF PRIMARY SITE OF INFECTION

Site	Children—35 hips	Adults—24 hips
Acetabular roof	11	1
Femoral neck	10	0
Synovial membrane ("Kissing lesions" in adults)	4	10
Undetermined	10	13

evidence was not conclusive, were discarded from this study. It may be of interest to note that among the eight unproved cases, seven had had the disease from five to twenty-six years. The remaining case in which the diagnosis was unproved was that of a boy of eight, with symptoms for seven months, who came from a tuberculous family and who died of miliary tuberculosis thirty-seven months after a fusion operation. Thus, nearly all the unproved cases are in the class of long-standing disease in which the repair process had progressed to an extent which made histological proof of the disease [6, 7] difficult to obtain.

The follow-up period ranged from one year and three months to seventeen years after operation, with the majority of cases having five to fifteen years of observation. The purpose of this study is to determine the comparative results of fusion in the two age groups and, in particular, the present status of early fusion in children aged five, or less,—representing more than 85 per cent. of the children in this series.

ANALYSIS OF DEATHS

There were six deaths, three in children and three in adults, constituting approximately 10 per cent. of the entire series (Table I).

It is obvious that the three deaths in the children's group could hardly be attributed to operative intervention, while the three deaths in the adult group might have been related to surgical interference.

THE PRIMARY SITE OF INFECTION

Serial roentgenograms of the diseased hips, taken at regular intervals from the onset of disease through a postoperative period—if the lesions were still discernible—were studied in an attempt to locate the primary site of infection. It must be stated, however, that no certain diagnosis can be claimed from these determinations. Where there was unmistakable roentgenographic evidence of a destructive lesion in one specific juxta-articular area to the exclusion of the other bones, within a reasonable limit of time, it was taken as the primary site of infection (Table II). Many of these determinations were corroborated at operation.

Among the undetermined cases were those of long-standing hip disease where the destruction had been so great as to leave no trace of identification of the initial process, while others were hips with multiple foci in the osseous structures. Of the four (Table II) in the children's group with primary synovial involvement, no bone lesions were discernible in the roentgenogram or at operation, but the pathological changes in the synovial membrane were such as to make the diagnosis of tuberculosis highly probable. The "kissing lesions", described by Phemister, were typical of the lesions in ten adults, presumably of synovial origin, although the so-called "kissing sequestra", probably a later phenomenon, were not demonstrated at this stage.

Tuberculosis of the greater trochanter has sometimes been implicated as an initial focus through which the hip joint may eventually become involved. During this period

there were three patients with lesions of the greater trochanter which were excised without the hips becoming affected. The acetabular-roof lesion, a decidedly common focus in hip-joint tuberculosis, may remain localized for a long period. Of considerable interest is a case with a destructive lesion in the lateral acetabular roof which was curetted and the cavity packed with bone chips. The hip has remained free and entirely without symp-

FIG. 1-A

Superolateral acetabular-roof lesion in a girl of six; except for a mild degree of coxa magna on the right the joint itself is not involved. Clinical manifestations: limp and pain; eleven months after onset.

FIG. 1-B

Appearance of the hip and acetabular roof six years after curettage of the roof lesion and implantation of bone chips, in lateral and anteroposterior views. The hip is entirely without symptoms.

to the present time, more than six years after the curettement (Figs. 1-A and 1-B). All these extra-articular lesions were confirmed as tuberculous. As the hip joints in these cases have remained free from disease, they are not included in this study.

Tuberculous coxitis resulting from an abscess originating in the vertebral column has been reported, although it is considered a rare complication. Hellstadius described a case in which autopsy confirmed a communication between the joint and the psoas abscess. In this series there were four cases of hip-joint disease in which clinical and roentgenographic evidence seemed to indicate such a course, though the exact mode of transmission could not be confirmed.

ANALYSIS OF HIP FUSIONS

Fusion operations were performed on fifty-six hips, thirty-three hips in children and twenty-three in adults, including an adult female with bilateral lesions. There were two cases of spontaneous fusion; both were cases of proved tuberculosis and in both there had been repeated surgical drainage. One case was in a girl of ten, when first seen in the Clinic, who had had a diseased hip since the age of two with persistent draining sinus from the beginning. Her hip became solidly fused when she was sixteen, with sinus draining intermittently for a few years following. The other case was found in an adult male of thirty-three, whose hip had become diseased at the age of six with persistent sinus since onset. No date can be set for the spontaneous fusion in his hip, for it was solidly fused when first seen in 1940. One sinus, however, was still open in the ischiorectal fossa when last seen in 1949.

The types of operative procedures used for primary fusions in this series are as follows:

Hibbs' extra-articular trochanteric graft	in 23 hips
Extra-articular graft with intra-articular chips	in 24 hips
Graft with complete intra-articular denudation and the insertion of chips	in 4 hips
Intra-articular denudation and the insertion of chips	in 3 hips
Ischiofemoral arthrodesis of Brittain	in 1 hip
Metal fixation with a Smith-Petersen nail	in 1 hip

The secondary procedures, largely supplementary in nature, did not usually follow a definite pattern of technique. In two instances, one in which an ischiofemoral arthrodesis was performed and one in which metal fixation with a Smith-Petersen nail was used, ankylosis was not secured.

Of the thirty-three hips in children operated upon, complete anatomical fusion was ultimately obtained in twenty-nine, while two children died and two others disappeared

TABLE III

SUCCESSFUL FUSIONS

Children			Adults	
Number of Operations Required	Age at Primary Fusion		Number of Operations Required	Cases
	5 or less (*Years*)	6 to 14 (*Years*)		
1 fusion	7 (39 per cent.)	6 (54.54 per cent.)	1 fusion	14
2 fusions	8 (61 per cent.)	3 (45.45 per cent.)	2 fusions	1
3 fusions	3	2	3 fusions	1
			4 fusions	1
Total cases	18	11		17

from the Clinic before fusion could have occurred. This represents successful fusion of 87.8 per cent: in children. Of the twenty-three hips in adults operated upon, seventeen are known to have been solidly fused. Of the remaining six, three patients died before fusion had taken place; two other hips had not fused in two years and in nine and one-half years, respectively, after operation; the fate of one remains unknown. Thus, in the adult group, the operative success is at least 74 per cent.

ANALYSIS OF SUCCESSFUL FUSIONS

The successful fusion of twenty-nine cases in the children and seventeen in the adults, and the number of fusion operations taken to achieve ankylosis in each, were closely scrutinized in regard to the patients' age at operation, the time intervals between the onset of the disease and the operation, and the time of the appearance of roentgenographic evidence of beginning fusion after operation. By roentgenographic evidence of beginning fusion is meant that there is shown a firm union of the graft to both the ilium and the femur, with early signs of intra-articular consolidation and of the arrest of the disease process. The interval between the appearance of consolidation and the final trabeculation varied from one to several years. The appearance of transarticular trabeculae is synonymous with the anatomical fusion referred to in this study. The children were divided into two groups according to the age of each child at the time of operation,—those under and those over five years of age (Table III).

It is to be noted that among the seven successful primary fusions in the younger group of children, two children were operated upon at the age of three, two at four, and three at five; while in the older group of children, the six children with successful primary fusion, two had an arthrodesis performed at six, one at eight, and three at eleven years of age. The percentage of successful primary fusions within the two groups of children is not significantly different. Combining the two groups, thirteen children had successful primary fusions, as compared with sixteen children who needed subsequent operations to obtain ankylosis. It is further noteworthy that of the eleven younger children who needed additional operations to achieve ankylosis, only five children were over five years of age by the time the final successful operative procedures had been performed,—one was ten, two were seven, and two were six years of age. The remaining six children were still five years old or less at the time the final operative procedures had succeeded in bringing about ankylosis. In other words, of the eighteen children aged five years or less when submitted to surgery, thirteen, or 72 per cent., had successful arthrodeses before they had reached five years of age. On the other hand, primary fusion operations seemed more successful in adults, as fourteen of seventeen adults, or 82 per cent., had successful primary fusion.

From an analysis of the time intervals between the onset of the disease and the primary operation, and between the primary and the secondary operations as related to the success of the fusion procedures and the time of the appearance of roentgenographic evidence of beginning fusion after operation, the following observations were recorded:

1. No difference was observed in the chances of the success of fusion procedures undertaken in children within seven to twenty-four months after the onset of the disease. A striking difference does exist, however, in the result of primary fusions performed within the first six months of onset, since the results were unsuccessful in five of seven patients operated upon in this period. Perhaps the term "unsuccessful" should be understood as being relative since, as will be seen later, some of the unsuccessful fusions in this series might have gone on to fusion had more time been allowed for fusion to take place before secondary procedures were undertaken.

2. Beginning evidence of fusion in the children seemed to take place between twelve and eighteen months after the fusion operation, with the average at the fifteenth month, in cases of primary fusion; between six and eighteen months, with the average at the

FIG. 2-A

A right femoral-neck lesion in a boy of three, two months after onset. The hip disease was clinically acute. Tissue culture was positive for tuberculosis.

twelfth month, in cases of secondary fusion. There was a trend toward earlier ankylosis following operation in the small group of cases in which operation was done five or more years after onset. While the time gained in effecting a fusion in this group is not spectacular, the resultant deformity of these hips, for which operation was delayed, leaves much to be desired, and was an indication that the operation should be done at as early an age as possible.

3. Ankylosis seemed to take place earlier after the fusion operation in the adult group than in children. The majority of cases of primary fusion in the adult group showed roentgenographic evidence of beginning fusion in from six months to a year.

FACTORS ACCOUNTING FOR THE SO-CALLED FAILURE OF PRIMARY FUSION

1. *Secondary Fusions Undertaken too Early*

In at least two cases in this series, re-fusion was advised, but fusion took place in the hips soon afterward without intervention. Secondary fusion procedures were performed within four to six months following the primary procedure in three cases, within twelve months in two cases, and within fifteen months in two cases, making seven

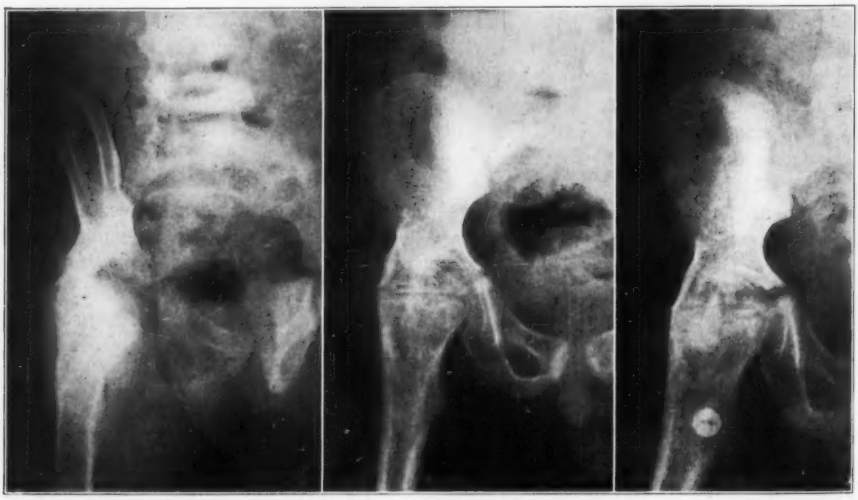

FIG. 2-B Fig. 2-C FIG. 2-D

Same hip one year, two years, and three years after Hibbs' extra-articular fusion, respectively, performed seven months after onset. Fig. 2-B shows the graft united with both ilium and femur. Figs. 2-C and 2-D show a progressive obliteration of joint space, a downward migration of the neck lesion, and a patent epiphyseal line. Growth was maintained.

cases in which the secondary procedure was carried out within fifteen months. If the fifteen-month period, which tallies closely with the time observed by Badgley and Hammond,

FIG. 2-E

Same hip five years later, showing almost complete fusion, a further migration downward of the neck focus, an intact epiphyseal plate, and growth maintained.

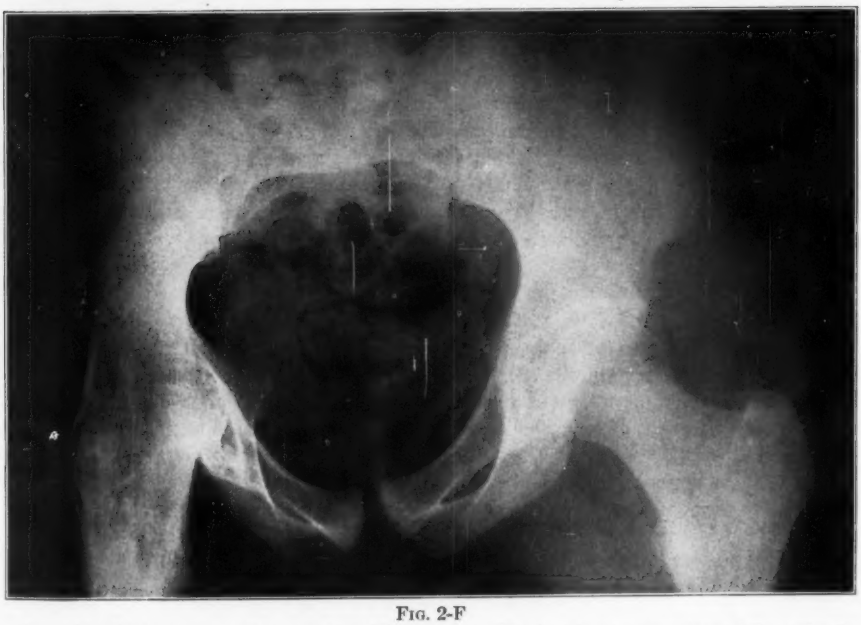

FIG. 2-F

Same hip thirteen years after one-stage Hibbs' fusion. The boy is now seventeen, fully active in sports. The shortening of the limb by clinical measurements is three-fourths of an inch; by scanograms, three-eighths of an inch.

is taken as an average time for fusion to occur after the primary operation, then fusion might have taken place in some of these seven cases without further operative interference. The fact that evidence of fusion occurred much earlier following second or third attempts at fusion in this series may indicate that, in some of these hips, consolidation might well have started when secondary procedures were undertaken.

2. *Inadequate Immobilization*

Insufficient cast immobilization, an insufficient total immobilization period, or too early ambulation may have been responsible for some of the failures. In re-checking the seven cases of successful primary fusion in children of five years of age or less, it was found that their average immobilization period in casts was from twelve to fifteen months, and the average time before ambulation was ten to twelve months. In most of the cases of unsuccessful fusion, on the other hand, the patients had an average immobilization period of from eight to twelve months and on the average began ambulation at the end of three months after operation.

Judging from roentgenographic evidence of beginning fusion in this series, it seems advisable to keep these children in adequate cast immobilization for twelve to eighteen months and to allow the beginning of ambulation no sooner than six months after operation, all to be guided, of course, by roentgenograms. In this Hospital, it is an established practice to bivalve the leg portion of the cast, leaving the anterior half continuous with the spica, to allow knee motion three months after operation. If fusion could be assured by prolonging cast immobilization for an additional few months, it certainly should be resorted to rather than to subject these patients to two or more fusion procedures. Roentgenographic evidence of fusion, following operative procedures in adults, seems to justify a correspondingly shorter immobilization period.

The inadequacy of immobilization may have another important bearing on the end results in many of those whose hips were ankylosed at a wide variance from the position originally intended. For instance, it was found that 60 per cent. of the ankylosed hips in children in this series had a flexion angle greater than 35 degrees, with varying degrees of abduction, adduction, external rotation, and internal rotation; while in the adult group the undesirable hip position amounted to a little more than 27 per cent. It is interesting to note the ankylosed position of the two cases of spontaneous fusion in this series: that of the girl was flexion of 40 degrees, adduction of 30 degrees, and internal rotation of 15 degrees; that of the man was flexion of 45 degrees, external rotation of 35 degrees, and neutral lateral position. Presumably, they had had inadequate and less sustained immobilization throughout the disease period.

3. *Miscellaneous Factors*

In five children and four adults the graft fractured after apparent union, which occurred in from seven to ten months after operation. Some of these fractures constituted indications for re-fusion. On the other hand, in at least two of the adult patients such graft fractures united spontaneously in three and nine months, respectively. There were five cases of graft absorption in children and two in adults, and in each group there were several failures of the graft uniting with either the femur or the ilium. They, too, constituted some of the cases requiring re-fusion. Whether or not early ambulation or inadequate immobilization, or both, could be held responsible for these unfortunate results is a matter of conjecture; but ultimate union of the graft to both the femur and the ilium after an initial period of non-union has been observed.

ABSCESS AND SINUS COMPLICATING HIP DISEASE

In two cases in each group, primary fusion operations were carried out in the presence of sinus. In one, an adult, the postoperative course presented complicated problems in

Synovial involvement of left hip in a boy of four, four months after onset. Note coxa magna and narrowed joint space. Tissue positive and guinea-pig test positive.

FIG. 3-A

coping with a secondary infection, necessitating repeated incisions and drainage. This hip was ultimately ankylosed after four attempts at fusion, with a total treatment period of more than eleven years. The other three cases were not extraordinary; sinus developed postoperatively; and healing took place after varying intervals. Sinus developed in very few of the children before operation, as they were operated upon before the sinus stage, while most of the sinus in the adult group had healed before the time of operation. In about 54 per cent. of the children and 48 per cent. of the adults in this series, sinus of varying extent developed postoperatively, and lasted from two months to several years, some of them intermittently.

THE STATUS OF EARLY FUSION IN YOUNG CHILDREN

Aside from the fact that 61 per cent. of the children of five years of age or less in this series required two or three fusion operations to achieve ankylosis, some of which were probably performed in anticipation of failure, and aside from the fact that a majority of the primary fusions in this group required, on the average, three to six months longer to become complete than did those in the adults, nothing in this study would seem to constitute a serious contra-indication to early fusion in young children. On the contrary, the array of evidence in favor of early fusion in young children appears so encouraging that it far outweighs the seemingly objectionable features of early fusion as enumerated, even when one takes the statistical figures in this study at their face value. The evidence is as follows:

1. Operative fusions were successful in 87.8 per cent. of the cases. This figure considers as failures the two deaths and the two patients whose fate yet remains unknown.

2. Of the eighteen children who were five years old or less when they underwent fusion operation, seven had successful fusions by the first attempt, eight by the second, and three by the third attempt. But in the eleven children who required additional procedures, six had their operations successfully completed while they were still five years of age or less, giving this age group a success rating of 72 per cent. in fusion operations.

3. There is unmistakable evidence in at least six cases in children, five of whom were below five years of age and one who was eight years of age at the time of operation, that the upper epiphyseal plate remained patent and growth seemed to have continued unhampered throughout the growth period, a fact observed also by Smith[8]. Two of these cases were followed for more than five years, one for six, one for thirteen, and two for

FIG. 3-B

Same hip thirteen months after Hibbs' fusion. Note firm union of the graft and patent epiphyseal plate.

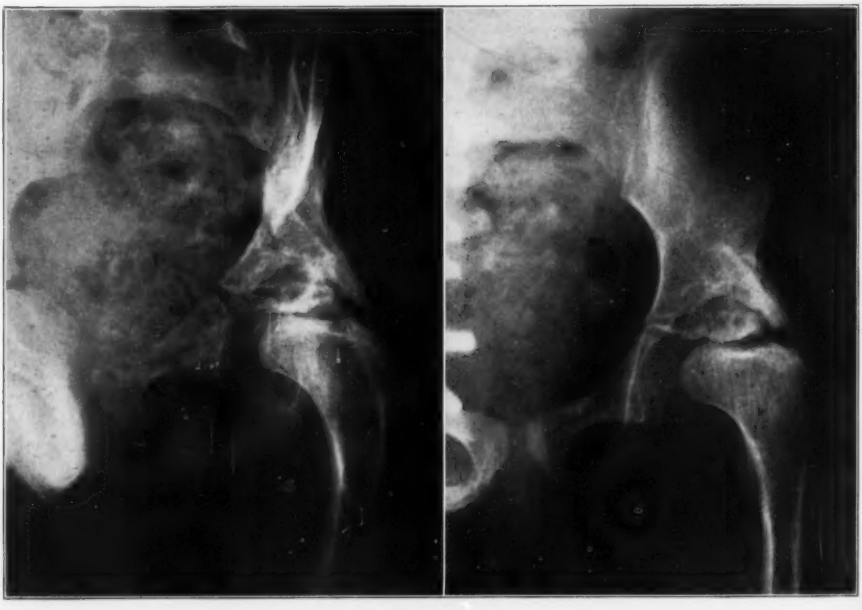

FIG. 3-C

Same hip sixteen and one-half months after fusion. Note a break of continuity of the graft at epiphyseal line (a common occurrence in children) due to growth, absorption, or fracture.

fifteen. Three had only one fusion operation, while the other three had two. Of the children having two operations, one was four, one was five, and the other was ten years old when

FIG. 3-D

Same hip six months after re-fusion (two years after primary fusion). Note the firm union of graft, obliteration of joint space, and patent epiphyseal line. Growth was maintained.

FIG. 3-E

Same hip in oblique and anteroposterior views four years and four months after re-fusion (five years and ten months after primary fusion). The boy is now ten years of age. Note the firm transarticular fusion and the patent epiphyseal line. Shortening by clinical measurement is one-fourth of an inch, by scanogram three-sixteenths of an inch.

the second operation was performed. The amount of shortening of the affected limbs at the end of the follow-up period was one-fourth of an inch in one, one-half an inch in two, three-fourths of an inch in two, and one and one-eighth inches in one. These are clinical measurements which are usually greater than the actual scanographic measurements because of the upward displacement of the head on the eroded acetabulum. Two representative cases of this group are here presented (Figs. 2-A to 2-F and 3-A to 3-E). It is noteworthy that, in about 50 per cent. of the children, the final shortening of the fused limb was within one to one and one-fourth inches, most of the shortening occurring in children five years of age or less when fusion operations were undertaken. In marked contrast, on the other hand, are the other 50 per cent. in whom greater shortening of the fused limb was encountered. These are the patients in whom invariably the disease had a longer duration before the operation.

4. Except for abscesses and sinus, equally prevalent in the older age group, there were no serious postoperative complications following early operative procedures in the younger age group.

Gill, in 1944, pointed out the danger of injury to growth centers about the knee followed by arrest of growth from long periods of immobilization in the treatment of this condition. He reported only ten cases of premature closure of the epiphyses at the knee in an analysis of 150 cases of tuberculosis of the hip in children treated at the New York Orthopaedic Dispensary and Hospital. This finding substantiated the validity of Hibbs' teaching of early fusion, early weight-bearing in a walking cast, and early mobilization in the treatment of this disease. McCarroll and Heath re-emphasized the advisability of early operative fixation to forestall the crippling effect of secondary changes in the extremity from prolonged immobilization.

It is beyond the scope of this study to consider the effect of secondary changes in the extremities of the affected side. Indeed, only one case of this nature was encountered in this study. It is not to be forgotten, however, that the extensive destruction at the hip joint and the complete loss of the upper femoral epiphysis in the patients for whom the operation was delayed in no small measure accounted for some of the limb-shortening in all patients.

CONCLUSIONS

1. A tuberculous hip rarely becomes fused of its own accord. The two cases of spontaneous fusion reported in this study had probably been transformed from an originally tuberculous infection to that of a predominantly pyogenic infection through long years of secondary invasion and repeated surgical drainages.

2. From the results of this study, it appears that there is added proof that early fusion in young children is amply justified. Conservative measures, aimed at building up the general health and causing the subsiding of acute symptoms, during the initial period of three to six months are necessary for a smooth postoperative course. In view of the present-day antibiotic therapy, even this short period of conservative treatment may be safely curtailed in some cases. In this way, the long recumbency in some forms of immobilization, and the distressing deformities, which usually follow long years of destruction from lack of internal fixation, may be avoided.

3. The average time for roentgenographic evidence of fusion to appear after arthrodesis in a hip in this series was fifteen months in children and twelve months in adults. Real anatomical fusion with transarticular trabeculae may take an additional one or more years. In the light of the present study, it seems advisable that no secondary fusion operations be undertaken before the fifteenth month in children or before the twelfth month in adults; that the average immobilization period be from twelve to eighteen months in children and from ten to twelve months in adults; and that the time for beginning ambulation be no sooner than the sixth month following arthrodesis in any case.

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GIANT-CELL TUMOR OF BONE

Current Status of Problems in Diagnosis and Treatment *

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Attention has recently been redirected to giant-cell tumor of bone through the publication of a symposium [3,9,21,24–26,29,30] dealing with certain aspects of its pathological changes and therapy, especially radiation therapy, and also of an editorial [8] concerned with the same subject. These expressions of opinion indicate that noteworthy progress has been made since the rather chaotic situation prevailing some twenty years ago, when no clear distinction was drawn between the so-called brown tumors of advanced hyperparathyroidism and the bona fide giant-cell tumors of surgical practice; when, collaterally, the neoplastic nature of giant-cell tumor of bone was open to question; when a wide variety of unrelated and generally less serious skeletal lesions were arbitrarily associated with giant-cell tumor in the guise of "variants"; and when, on the whole, no general appreciation of the seriousness of certain giant-cell tumors of bone existed.

On the other hand, it seems equally clear from the articles cited and from many others in the recent literature that some of the pertinent advances of the past ten years have not as yet been fully assimilated and that a good deal of uncertainty still prevails in regard to the clinical and pathological recognition of genuine giant-cell tumor, which must be dispelled before observations in regard to the efficacy of treatment are to have much significance. Stated more explicitly, people must be talking about the same thing before their results can be compared.

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In 1940, Jaffe, Lichtenstein, and Portis attempted to define what should properly be regarded as genuine giant-cell tumor, as a basis for more accurate diagnosis and rational therapy. In this survey, giant-cell tumor of bone was interpreted as a distinctive neoplasm arising apparently from the (non-bone-forming) supporting connective tissue of the marrow, which could be readily identified on the basis of its cytological details. Specifically, it was stated that the tumor was composed of a vascularized network of spindle-shaped or ovoid stromal cells, regularly and rather heavily interspersed with multinuclear cells (apparently syncytial stromal cells) as an integral part of the cytological pattern. As an important corollary of this study, it became evident that genuine giant-cell tumor should be completely divorced from its spurious "variants" with which it had been, and to some extent is still, confused. As Jaffe [12] has aptly remarked, these so-called "variants" have little in common clinically and anatomically with genuine giant-cell tumor, and also little in common with each other except an auspicious prognosis.

As a result of subsequent detailed investigation of these alleged variants of giant-cell tumor, there evolved a clearer concept of these lesions as distinct clinical and pathological entities in their own right. Thus, the so-called spindle-cell or healing or xanthic variant was superseded by the rather common lesion now called non-osteogenic fibroma of bone [13]. The "calcifying or chondromatous giant cell tumor" described by Kolodny, Ewing, and Codman was reinterpreted and designated benign chondroblastoma of bone [14,23], in the belief that it represents an independent benign tumor of cartilage-forming connective-tissue derivation, which is unrelated histogenetically to giant-cell tumor. Cognizance should be taken of the opinion of Willis [28] that a cartilaginous variant of genuine osteoclastoma does exist, although his dissent appeared to be based essentially upon inferences gleaned from a single specimen (labeled "chondromatous osteoclastoma") which the writer would regard unequivocally as an instance of chondromyxoid fibroma of bone [15, 20]. Recently, still another, rather common pathological entity frequently mistaken for giant-cell tumor and referred to generally as "atypical, subperiosteal giant-cell tumors in unusual locations", has been clearly delineated under the designation of aneurysmal bone cyst[22]. There now appears to be general agreement that the so-called giant-cell tumors of synovial or tenosynovial tissues are wholly unrelated to giant-cell tumor of bone. They appear to represent peculiar granulomata rather than true neoplasms, and the condition has been described as such by Jaffe, Lichtenstein, and Sutro under the head of pigmented villonodular synovitis, bursitis, and tenosynovitis.

Much of this confusion in pathological diagnosis stems from the circumstance that the common phagocyte or macrophage of skeletal tissues, and particularly of bone, happens also to be a multinuclear cell. As is well known, these multinuclear cells make their appearance at sites of resorption and reconstruction of bone or cartilage, organization of hemorrhage or inordinate vascularity, reaction to degeneration and necrosis of tissue, and repair generally. Thus, virtually any skeletal lesion presenting an appreciable number of such multinuclear phagocytes, for whatever reason, may come to simulate giant-cell tumor in certain fields, although usually not throughout, so that some discrimination may be required for the distinction to be made with assurance. It is unfortunately true that many lesions are still mislabeled giant-cell tumor by pathologists with limited experience in bone pathology, mainly because they present a scattering of multinuclear cells. The writer does not intend to imply, however, that the distinction is invariably simple, particularly if one is guided by the cytological picture alone, without taking into consideration the location of the lesion, the age of the patient, the history, and other relevant clinical features. For example, one may on occasion observe conspicuous giant-cell reaction to what appears to be massive intramedullary hemorrhage in the shaft of a long bone, that may be quite suggestive of giant-cell tumor.

In regard to significant clinical inferences, our observations led us to conclude that genuine giant-cell tumor, in contrast to some of the alleged variant lesions, is seldom

observed in patients below the age of twenty and that, when situated in a long bone, it usually involves the end of the bone. Willis [29] and some of his British colleagues [3] are inclined to question the validity of this view, but it is the writer's impression that they are not so critical as they might be in accepting, as giant-cell tumors, lesions that develop in the metaphyses of young patients. As noted, such lesions, in the great majority of cases, prove on close histological scrutiny to be instances of other conditions, on the whole less serious than giant-cell tumor (non-osteogenic fibroma, bone cyst, chondromyxoid fibroma, et cetera). Incidentally, the statistical data cited editorially [8], apparently culled from the comparatively old article by Christensen, likewise give one a somewhat distorted impression of the incidence and localization of giant-cell tumor, particularly in the bones of the upper limb, the skull, and the pelvis and vertebrae, where, as has been indicated elsewhere [22], aneurysmal bone cyst is prone to develop.

If considerable emphasis has been placed upon the foregoing considerations, it is because they have an important practical bearing on the treatment and prognosis of giant-cell tumor. Until there is general agreement, in actual practice as well as theory, as to what constitutes giant-cell tumor, it seems futile to discuss results of treatment by one method or another,—treatment of what, one may ask. Stated more explicitly, if the real giant-cell tumors are freely watered down by other lesions of no serious consequence, then results of treatment by any method can be made to look good.

To cite a relevant and by no means isolated instance, a paper by Leucutia and his associates several years ago purported to show the gratifying effectiveness of irradiation in the treatment of thirty-three giant-cell tumors. Analysis of their data revealed, however, that some of the alleged giant-cell tumors represented epulides, others were observed in children and adolescents, and still others were located in the shafts of long bones. Of the total number presented, there were approximately seven tumors in the ends of limb bones of adults which the writer would regard as probable giant-cell tumors; of these, four came to amputation and two ultimately proved fatal. To cite another instance in point, a paper by Buschke and Cantril recently appeared, likewise advocating the use of roentgenotherapy on the strength of satisfactory clinical results obtained in ten cases, scarcely one of which could be accepted as a convincing giant-cell tumor. It is pertinent to quote the revealing admission of Cade that "central non-trabeculated cystic tumours in the classical sites at the upper end of the tibia and lower end of the femur respond badly to radiation whereas [expanded] trabeculated tumours with 'soap bubble appearance' [in the flat bones of the pelvis and shoulder girdle and particularly in the vertebrae] respond well, especially if they have not already been curetted". The former appear to represent genuine giant-cell tumors, the latter aneurysmal bone cysts in all probability.

On the other hand, if one adheres to a strict definition of what should be regarded as giant-cell tumor and strips away all of the alleged variants, then what is left constitutes a formidable neoplasm to be reckoned with under any circumstances. It has become increasingly evident that, while giant-cell tumors are not necessarily "sarcomata", neither are they all "benign", and that, with respect to potential seriousness, they may run the whole gamut from one extreme to the other. There appears to be substantial agreement, among pathologists at least, that, while many giant-cell tumors are successfully treated by thorough curettement or irradiation, some are undoubtedly aggressive and prone to recur; occasional ones behave like frank sarcomata, either when initially observed or, more often, after one or more local recurrences [27]. Obviously, statistically valid as well as accurate data reflecting appropriately long follow-up records are not readily collected by any single observer. It is the writer's impression, however, as a working hypothesis, that, given a sizable group of proved giant-cell tumors, approximately one-half are likely to have a favorable outcome if properly treated by whatever method, approximately one-third are likely to prove more aggressive and recur after treatment (and a considerable proportion of these may eventually come to amputation), while the remaining 15 per cent., more or less, will

be frankly malignant and prone to metastasize to the lungs. An occasional giant-cell tumor will be found to be malignant at the time of initial tissue examination, but more often one has to reckon with malignant change incidental to one or more local recurrences.

Along these lines, as a guide to therapy and with a view to forecasting prognosis within certain limits, Jaffe, Lichtenstein, and Portis recommended that giant-cell tumors be subclassified into three grades, arbitrarily designated numerically as I, II, and III, and showing respectively insignificant, moderate, and pronounced atypism of their stromal cells. Willis [29] and Russell have recently contended that the giant-cell tumors which prove to be aggressive or eventually frankly malignant are not initially distinguishable from their benign fellows. One must be wary, however, of making broad generalizations from isolated or limited observations, and the writer can only reiterate that, while no method of appraising potential aggressiveness from cytological criteria is infallible, in his own experience the grading of giant-cell tumors has proved to be of practical value. Specifically, the giant-cell tumors which pursued an aggressive clinical course were, for the most part, tumors of Grade II rather than Grade I initially, while those that eventually became frankly malignant (Grade III) likewise revealed a portent of this ominous tendency in previous tissue specimens.

PROBLEMS IN THERAPY

In regard to therapy generally, one finds increasing acceptance, in theory at least, of the basic concept that roentgenographic interpretation alone may be fallacious and that rational treatment must be predicated upon reliable pathological diagnosis, regardless of whether one favors surgery or roentgen irradiation as the treatment of choice. In the main, however, recent contributors have not been greatly concerned with pathological analysis of their material, but have attempted to point out the advantages of roentgenotherapy over surgical treatment, or vice versa. Most of them agree that it does not seem to be a good plan to combine the two methods, although precisely why is not clearly indicated. Beyond that, there has been no comprehensive survey of specific problems entailed in the treatment by either method of a proved giant-cell tumor that is approached for the first time, or of one that has already recurred after treatment.

It would seem to the writer that a clear statement of these problems, without prejudice in favor of either modality, is the first essential step in the planning of well controlled, long-range clinical experiments that alone can supply the data upon which a sound therapeutic program must eventually be based. It is one of the main purposes of this paper to attempt to outline some of these problems.

As indicated, biopsy should be resorted to before treatment is instituted, irrespective of the method of treatment employed. If one favors surgery, the diagnosis can be established by conservative but adequate surgical exposure (avoiding pathological fracture of the attenuated cortex) and frozen section of a bit of representative tissue. There should be no difficulty on frozen section in identifying a giant-cell tumor and in ruling out a sarcoma, although the grading of the tumor may require good paraffin slides. The tumor may then be treated by excision, resection, or thorough curettement, depending upon its location and size.

Whenever it is possible to excise or resect the tumor *in toto*, this should be the method selected as the most certain and quickest way to effect a cure. Unfortunately, the majority of giant-cell tumors present as relatively large growths in the lower end of the femur or the upper end of the tibia, where resection as a routine procedure is hardly feasible. For these, thorough curettement is the procedure generally followed by those committed to surgery. In cases treated successfully by this method, one wonders what happens to residual bits of tumor tissue adhering to the walls of the cavity created by curettement, and can only surmise that they probably undergo necrosis as a result of interference with their blood supply. Along the same line of thought, is anything gained by chemical cauterization of

the walls of the cavity, employing zinc chloride, for example, or does this merely impede osseous reconstruction? Also, is packing of the cavity with bone chips, with a view to hastening osteogenesis, desirable as a routine measure? While most surgeons would answer this latter question in the affirmative, their impression is likely to be largely subjective.

In regard to the desirability of employing supplementary irradiation directed against any residual tumor that may be present, the statement has been made[8]: "Irradiation as an adjuvant to curettage may apparently stimulate recurrence or increase the aggressiveness of the lesion". While this contention undoubtedly echoes a large section of current opinion, actually what proof is there of its validity, and, if the suggested hazard is real, does it apply to small or moderate, as well as large, doses of roentgen ray? Still another suggested approach about which one must keep an open mind is the use of limited x-ray irradiation prior to curettement, as advocated by Haggart and Hare. These authors claimed that such preliminary irradiation, adequate to arrest tumor growth temporarily, yet not intense enough to induce bone necrosis or prevent soft-tissue healing, renders the tumor tissue sufficiently firm and avascular to permit thorough curettement.

If one is committed to x-ray irradiation of giant-cell tumor by choice, sufficient tissue for diagnosis can still be obtained by resorting to aspiration or drill biopsy, repeated if necessary in the event that the initial specimen is inadequate or the findings inconclusive. Most roentgen therapists freely admit, off the record at least, that roentgenographic interpretation alone has inherent limitations, in spite of Brailsford's opinion to the contrary, and that accurate pathological diagnosis prior to treatment is eminently desirable. They readily concede, on the one hand, that bone lesions less serious than giant-cell tumors may simulate the latter roentgenographically, and, on the other hand, that occasionally malignant tumors of one kind or another may not be readily distinguishable from giant-cell tumor if one has to rely solely on the roentgenogram. Unfortunately, some of these roentgen therapists appear content to pay lip service to the principle involved; others resort to biopsies, but are willing to accept casual impressions, being themselves unaware of the subtleties in differential diagnosis which confront the pathologist. There seems to be good reason to hold empirically that x-ray irradiation *per se* is capable of effectively controlling a certain number of giant-cell tumors, particularly the less aggressive ones. In the absence, however, of acceptable proof of the pathological identity of the lesions treated, claims for the efficacy of such treatment lack scientific validity despite long-range follow-up observations. To the writer's knowledge, a well controlled and altogether convincing sizable series of giant-cell tumors treated by roentgenotherapy and followed for five to ten years has yet to be presented.

This tendency to casual empiricism is further reflected in the fact that no unanimity of opinion seems to exist among the exponents of roentgenotherapy in regard to the optimum tumor dose that should be employed or the manner in which it should be administered. Thus, some contend that a total tumor dose as small as 1,000 r suffices to effect a cure; others administer as much as 2,000 or 3,000 r; while still others employ doses considerably in excess of 3,000 r, in spite of the demonstrated hazard that some patients so treated may manifest the development of sarcoma in the lesion following a latent interval of five or more years after irradiation. The writer has personal knowledge of several cases in which sarcoma developed within an irradiated and ostensibly cured giant-cell tumor after a long latent interval, and Cahan, Woodard, Higinbotham, Stewart, and Coley have reported several others in connection with a general survey of sarcoma in irradiated bone. Such cases are not very common in relation to the total number irradiated, but they are sufficiently numerous to indicate that bone lesions cannot be irradiated with very large doses of roentgen ray with impunity. The practical importance of these observations is that, if one elects to irradiate a giant-cell tumor, one should use the smallest possible effective tumor dose,—that is, if 1,000 r will suffice, one should avoid using 3,000 r or more.

The effective control of a giant-cell tumor which has already recurred locally following

treatment, whether curettement or irradiation, raises still another set of problems that require clarification. In the event that the initial therapy has been surgical, what is the likelihood that additional curettement will effect a cure? If one is tempted to employ irradiation rather than further surgery, one runs counter to the popular admonition that combined treatment leads to generally unsatisfactory results. It may be true that roentgenotherapy for a tumor which has recurred following curettement is not so effective as one might wish, but are the failures necessarily attributable to combined treatment? May they not indicate merely that irradiation is not nearly so effective against the aggressive giant-cell tumors of Grade II as it is against the more favorable tumors of Grade I? Before categorically condemning combined therapy in such cases, one must, in all fairness, compare the follow-up results with those in which re-curettement has been employed for recurrence.

In the event that the recurrent tumor has been initially treated by roentgenotherapy in presumably adequate dosage, what is the likelihood that a second course of irradiation to the limit of tolerance will be any more effective? In so far as the feasibility of supplementary surgery is concerned, one can readily appreciate the hazards of failure of tissue healing and of infection entailed in the curettement of irradiated tissues, but are these necessarily a deterrent in dealing with tumors which have received only moderate doses of roentgen ray, and precisely what is the limit of tolerance? If neither of these alternatives seems attractive, should one advocate surgical ablation for giant-cell tumors that have recurred in spite of what seemed to be adequate irradiation? As Coley has remarked: "There are few situations in the entire field of bone tumors which afford greater anxiety or which tax the judgment and intuition of the professional adviser more sorely than these cases of giant cell tumor which fail to yield to radiation therapy". The writer does not presume to have the answers to the questions raised, but submits that, notwithstanding all that has been written on the subject of giant-cell tumor, there are still basic problems in therapy which require renewed consideration and cannot be circumvented by facile generalizations.

In dealing with a giant-cell tumor which has recurred more than once, the writer is firmly of the opinion that ablation should be resorted to, if the tumor is in a surgically accessible site, since to temporize further is to invite frank malignant change and pulmonary metastasis. In reading the published case reports on malignant giant-cell tumors, one cannot fail to be impressed by the fact that amputation was much too late to be effective.

It seems highly improbable that the problems in therapy indicated can be resolved on a broad front by the accrued observations pertaining to single cases or even small groups of cases. Also, inasmuch as no single institution, large though it may be, can hope to accumulate sufficient data to be conclusive (except perhaps over a twenty-year span), it might be mutually profitable for a number of participating hospitals to set up a giant-cell-tumor registry. The registry could accept responsibility for establishing accurate pathological diagnoses, standardizing therapeutic procedures, and recording follow-up observations extending over a period of five to ten years or more, whenever possible. In this way, it would be entirely feasible to conduct a well planned and controlled, large-scale clinical experiment bearing on the relative efficacy of surgical treatment, x-ray irradiation, or both combined. Thus, if the comparative end results of each method were known in, let us say, fifty or more proved cases of giant-cell tumor, one would have a sound foundation for specific recommendations in regard to treatment. Needless to say, this approach would also yield much information of fundamental importance concerning the biological behavior of giant-cell tumor.

SUMMARY

The uncertainty that still prevails in many quarters as to what should properly be regarded as giant-cell tumor of bone (stripped of its spurious variants) has had the effect

of vitiating most of the contributions to the literature of the past ten years. This confusion must be dispelled before reports in regard to the efficacy of treatment, by one method or another, are to have much significance. To this end, the writer has outlined some of the pertinent advances of the past decade concerning the clinical and pathological features of genuine giant-cell tumor and of certain other lesions which are frequently mistaken for it. The necessity for establishing accurate pathological diagnosis before instituting treatment, whether surgical or irradiation, has again been emphasized.

In regard to therapy, the writer has tried to survey some of the specific problems entailed in the treatment, by either method, of a giant-cell tumor that is approached for the first time, and of one that has already recurred after treatment. Significant progress in the solution of these problems can only come about through the effective planning, possibly through the agency of a giant-cell-tumor registry, of well controlled, long-range clinical experiments that alone can supply the data upon which a sound therapeutic program must eventually be based.

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ADDENDUM

Since this paper was submitted for publication, the author has seen the article by Leucutia and Cook, who have taken cognizance of the fact that sarcomatous change occurs in a significant number of irradiated giant-cell tumors. Leucutia and Cook[c] were inclined, however, to regard this as a "natural sequence of events unaffected by the type of treatment given". This ostrich attitude was reiterated in an editorial published in the *American Journal of Roentgenology* in December 1949 [b], in which it was stated that ordinary doses of roentgenotherapy cannot be held responsible "for any sarcomatous induction or conversion. It is more likely that osteogenic sarcomas which develop in giant cell tumors treated by roentgen therapy or arise after many years at the sites of giant cell tumors apparently cured by ordinary doses of roentgen rays represent samples of malignant neoplasia occurring during the natural sequence of events."

No one will deny that some of the neoplasms in question were injudiciously over-irradiated with very large doses or that, on the other hand, giant-cell tumor may undergo malignant change, either spontaneously without previous irradiation or following surgical treatment alone. However, as Stewart [d] has pointed out, there appear to be distinct cytological differences between spontaneous malignant giant-cell tumors and sarcomata induced by irradiation of giant-cell tumors. Further, the point raised by Cahan [a] seems well taken,—namely, that it usually requires five years or more for an irradiation-induced sarcoma to develop, whereas a spontaneously developing malignant giant-cell tumor may be reasonably expected to manifest itself within a considerably shorter interval. These points are raised not by way of engendering controversy, but rather to indicate that those who hold roentgenotherapy responsible for a certain number of instances of malignant transformation in giant-cell tumor have valid arguments which cannot be summarily dismissed.

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b. EDITORIAL: Is Roentgen Therapy Hazardous in Benign Bone Lesions? Am. J. Roentgenol., **62**: 891–893, 1949.
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RESECTION OF DORSAL VERTEBRAE IN CONGENITAL SCOLIOSIS*

BY PHILIP WILES, M.S., LONDON, ENGLAND

Congenital defects of the vertebrae are a tempting morsel for the surgeon. Not so much the dreadful multiple defects in which the whole spine has gone awry, but rather those with a single hemivertebra waiting to be picked out like a chestnut roasting between the bars of a grid. The honor of being the first to accomplish this feat falls to von Lackum who, in 1924, successfully enucleated the eleventh dorsal vertebra, and, a few months later, part of the tenth dorsal from the same patient. This was the first operation in a series of ten which were reported after his death by Alan DeForest Smith. Other successes were recorded by Royle in 1928 and by Compere in 1932. These operations were all enucleations of a single lumbar or low dorsal vertebra and were performed for both congenital and acquired defects. There have, however, been no reports since that time and the operation has apparently fallen into disuse, it may be because the end results were not sufficiently encouraging.

I decided to tackle the problem in rather a different way and perform a wedge resection instead of an enucleation; a patient with multiple deformities, who could not easily be made worse, was to be selected for the first attempt. The whole affected area would be excised, and also the adjacent epiphyses of the vertebrae on each side of it. Cancellous surfaces of bone would then be left in apposition and these might be expected to unite readily, forming a single, composite vertebra of fairly normal shape. It seemed advantageous to try first the mid-dorsal region, because at this level there would be reasonable stability after transection of the spinal column. The technical difficulties of the operation, aided by modern methods of anaesthesia and transfusion, were not too great. I have, however, performed it only twice, because in both instances a kyphosis subsequently developed and proved to be an even worse deformity than the original scoliosis was likely to have become. These two cases are reported now partly in the hope that some method of preventing the kyphosis will be forthcoming, and partly to warn others against making a similar attempt before this difficulty has been overcome.

CASE 1. L.D. was first seen when fourteen days old. The deformity was so severe as to be obvious at that age, and the parents agreed to operation even after the risks had been explained. (The best age for the attempt seemed to be about twelve months, because by then there is a very considerable resistance, and yet the whole body is still so plastic that it can adapt itself to almost any insult.) By the time of operation the lateral angulation of the spine had increased from 45 to 60 degrees, the condition being shown in Figure 1-A (left).

The anaesthetist plays almost as large a part as the surgeon in such an operation as this, and I was fortunate enough to secure the help of Dr. F. W. Roberts. He set up a blood drip and used only pentothal introduced into the drip. This is thought to be the first recorded occasion of continuous intravenous anaesthesia in so young an infant.

The operation was performed in stages. At the first (May 3, 1941) the entire neural arch of the affected vertebrae, including the pedicles, was resected. In addition, the transverse processes and the heads and part of the necks of the ribs on the concave side of the curve were excised, because it was thought this would make it easier to stretch the tissues on the concave side, always a difficult problem. At the second stage, a week later, the first incision was reopened and a new one was made in the line of the tenth rib on the convex side. Except for the skin incision, all cutting of soft tissues was done with diathermy. Three ribs were resected subperiosteally and the transverse processes were removed; two neurovascular bundles were deliberately divided, and the erector spinae were cut transversely. There was then no difficulty in exposing the bodies of the vertebrae both laterally and anteriorly, but some care was necessary because the meninges, the pleura, and the aorta were all pulsating at various depths in the wound. A wedge of bone and intervertebral discs, having a base about one and one-quarter inches thick, was excised with "nibblers", knife, and chisel. It included three vertebrae, dorsal eight, nine, and ten, or at any rate all there was of them, and the adjacent

* Abbreviated from a paper read at the Annual Meeting of The American Orthopaedic Association, Virginia Beach, Virginia, May 26, 1950.

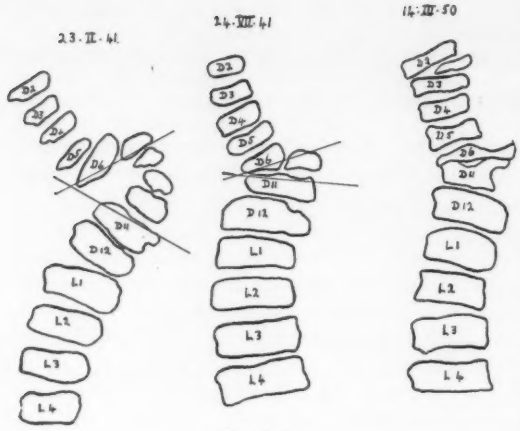

FIG. 1-A

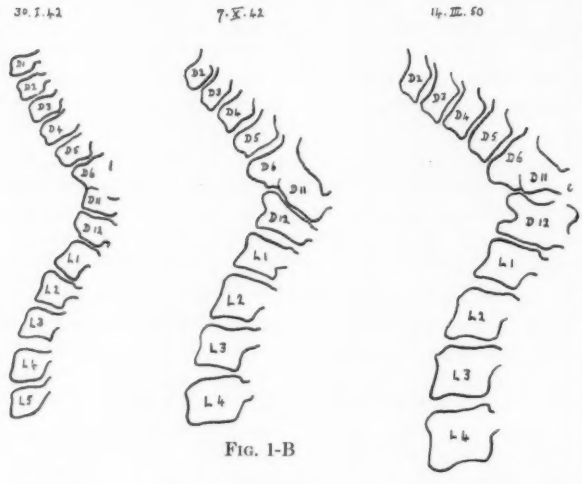

FIG. 1-B

surfaces of dorsal seven and eleven. There was a mass of cartilage on the concave side which could not be completely removed, and therefore it was impossible to arrange that the apposed surfaces consist of bone only. It was interesting, when cutting through the middle of a disc, to see the nucleus pulposus being rapidly extruded. The spine was not grossly unstable in spite of being transected, and the gap could not be completely closed by lateral bending. A little more bone and cartilage were removed from the concave side, but complete closure was still impossible. The wound was therefore sutured and a plaster cast, including the trunk, one leg, one arm, and the head, was applied. There was almost no bleeding throughout the operation, except from bone. The time taken was one and one-half hours; 5.5 ounces of blood was infused during the operation.

A few days later the plaster was cut around the middle, and hinges and a turnbuckle were fixed in the fashion of a Risser jacket. The cast was removed after three months and, although the back appeared clinically to be straight, the roentgenograms still showed an appreciable deformity (Fig. 1-A, center). An abnormal vertebral body was still present at the site of resection and this clearly had to be removed. Therefore, on August 9, 1941, the old wound was reopened; the exposure was rather harder than before because of scar tissue. A further wedge of bone, having a base half an inch thick, was removed; it included the remaining rudimentary vertebra. The effect of this operation was to produce more, but not full, correction of the spine (Fig. 1-A, right).

The fate of the abdominal muscles supplied by the two severed intercostal nerves is of interest. At first there was a bulge of the lower abdominal wall which had to be supported by a corset; but by the time the child was six years of age there was no detectable weakness and it seemed almost as if the muscles had acquired a fresh nerve supply.

The original roentgenograms, which were not made in easy circumstances, are hard enough to interpret and reproductions would be valueless; tracings have, therefore, been prepared. Figure 1-A shows that four dorsal vertebrae, the seventh, eighth, ninth, and tenth, were removed and the spine, as viewed from the front, has been greatly straightened. Another hemivertebra has appeared between dorsal two and three.

The kyphosis began to develop within six months of operation, whilst a plaster jacket was still being worn. No preoperative lateral roentgenograms are available, but the progress is clearly shown in Figure 1-B. The child is now ten years old, both mentally and physically healthy, but the deformity is extremely severe.

A second trial of the procedure was delayed by the War until May 30, 1946.

CASE 2, R.L. The patient was aged fifteen months at the time of operation, and he, also, had severe multiple defects (Fig. 2-A, left). Dr. Bernard Johnson gave the anaesthetic; he preferred gas and oxygen to pentothal and it proved to be equally satisfactory.

The operation was performed in stages as before. At the first stage the neural arch and pedicles were removed, and also the transverse processes and the heads and necks of the ribs on the concave side. A week later the posterior thirds of the sixth, seventh, and eighth ribs on the convex side were resected subperiosteally, and a wedge with a base about one inch thick was removed from the vertebral column. It included the rudimentary sixth and seventh dorsal vertebrae and the adjacent epiphyses of the fifth and eighth. There was no greater technical difficulty than before, and it was found unnecessary to divide the neurovascular bundles. On this occasion, however, there was a postoperative fever of hectic type, the diagnosis of which baffled everybody until in desperation the cast was removed; it was found that the infant had rubbed a sore on the back of the scalp where it was in contact with the plaster and an extensive subaponeurotic abscess had developed.

The immediate correction of the lateral curve was encouraging, and a year after operation the infant was walking and playing normally. However, the kyphosis began to develop only too quickly (Fig. 2-B), and

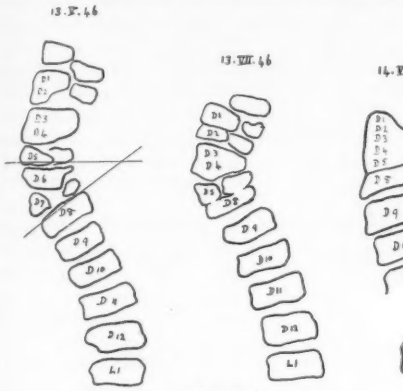

FIG. 2-A

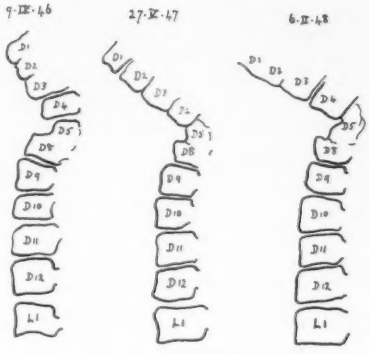

FIG. 2-B

soon there were signs of commencing spinal-cord pressure. The paraplegia progressed slowly and inexorably, but because of a series of intercurrent illnesses, exploration of the cord was impossible for more than a year; then it was too late to do any good. The child died last year, three years after operation. Unfortunately, we did not learn of his death in time to request a postmortem examination.

If anything has been achieved by these operations, it is to show that dorsal vertebrae, almost as many as is wished, can be removed from infants. Great possibilities are opened up, if only the subsequent development of a severe kyphosis can be prevented. The cause is uncertain. One factor, which is clearly shown in the second patient, is that the remaining vertebrae are themselves defective. This would not be so in the case of a single hemivertebra, but one could not at present have the temerity to attempt the operation when the potential deformity is comparatively slight. The major difficulty appears to be that resection of one face of the two vertebrae which are to be apposed interferes with their subsequent development, even though intact growth discs remain at the top and bottom of the new, composite vertebra. It is, therefore, doubtful if the deformity would be prevented by spine fusion, even should it be practicable at such an age.

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DISCUSSION

DR. EDWARD L. COMPERE, CHICAGO, ILLINOIS: Nineteen years ago, I became very much interested in the problem of trying to correct, in children still at a tender age, curvatures of the spine that were caused by hemivertebrae. At that time I could not find any recorded description of an operation of this kind. Von Lackum's work had not been reported, and I believe that some of his patients, if not all, were older. I did find that Codivilla[1], a brilliant young Italian surgeon, had suggested in 1901 that it should not be too dif-

1. CODIVILLA, A.: Sulla scoliosi congenita. Arch. di Ortop., **18**: 65–78, 1901.

ficult to excise a hemivertebra and correct a curvature in infancy. The professor who had charge of the orthopaedic service refused to permit him to carry out the operation. Also, Royle mentioned that he had excised a hemivertebra. He did not mention the end results.

Of the two patients whom I operated upon, one was thirteen months of age at the time of admission to the hospital, and the other was sixteen months. In the first, I was able to remove two hemivertebrae. Unlike Mr. Wiles, I did not attempt to excise the adjacent intervertebral disc or a portion of the adjacent vertebral bodies. I shelled out each hemivertebra, leaving small fragments of bone all the way around; it was then possible to collapse the hollowed-out space. In the first patient, I removed the hemivertebrae from the level of the eleventh thoracic and first lumbar vertebrae. Two ribs, which had been removed to obtain the exposure, were placed on the laminae for fusion of the spine.

The second patient had one large hemivertebra at the level of the first lumbar. This was removed, the disc being left intact on each side. The correction of the curvature was not complete, but there was marked improvement. I didn't know about the Nachlas staples at that time, but I did use a tibial bone graft and inlaid it in the vertebral bodies from the twelfth thoracic to, and including, the third lumbar on the convex side. This second patient had the better result of the two for the short time that he lived. He became normally active, running, jumping, or climbing. The family moved to Wisconsin and, about eighteen months after operation, I received a letter stating that the child had died of lobar pneumonia. A kyphosis had not developed. In Case 1 a severely deforming kyphosis did develop, similar to those shown by Mr. Wiles. Had that not been the case, I would suspect that the reason for the kyphosis in Mr. Wiles's two patients may have been the wide excision not only of intervertebral discs, but of the cartilage plate itself, from adjacent vertebral bodies above and below the hemivertebrae which he had excised.

Height growth of a vertebral body is produced by newly formed cells laid down by the vertebral cartilage plate. This plate is to the vertebral body what the articular cartilage at the end of a long bone is to the epiphysis. The epiphysis grows, not from the epiphyseal line, but from the joint side, and the vertebral bodies grow in height from this cartilage plate. Undoubtedly that could account for the failure of these vertebrae to grow. However, in my first patient I did not damage the cartilage plates, yet marked kyphosis developed.

During the last eighteen years, I have received letters from time to time, asking whether or not I still advise the operation. I did not and do not recommend it. Fusion of the spine at an appropriate age would be a safer operation and should afford as good an end result. Soon we may all be using Nachlas's staples on the convex side to check and hold back growth and correct the spine curvatures.

I wish to thank Mr. Wiles and to congratulate him for a most interesting and courageous presentation.

DR. ALAN DEFOREST SMITH, NEW YORK, N. Y.: I am particularly interested in this paper because, as Mr. Wiles stated, Dr. von Lackum and I operated on such a patient twenty-six years ago, removing a hemivertebra from the lumbar region. That patient is now thirty-two years of age and has an excellent result with a straight spine. It is not difficult to remove a hemivertebra in the lumbar region. We did it in two stages, using the posterolateral approach that Royle had employed for sympathectomy in removing the body of the hemivertebra in the first stage; in the last stage, a few weeks later, we removed the posterior arch and did a spine fusion. This was the technique used in the later cases. We were sufficiently encouraged by the results in the first case to repeat the operation. In only one case was the operation done in the thoracic region. That was in an older girl who had had spine fusion for ordinary idiopathic scoliosis; the fusion had left her in a poor position, so that she was off balance and quite badly deformed. A wedge-shaped osteotomy was done in the bodies on the convex side, the corresponding posterior margins were removed, and fusion was carried out. She got a good result and kyphosis did not develop.

However, in several of the other cases in which a hemivertebra had been removed in the lumbar region, severe kyphosis did develop, in spite of the fact that a spine fusion had been done. That discouraged us from going on with these operations, and we have not done any since that time.

In the two cases that Mr. Wiles presented, the condition was very severe and obviously the patients were going to have disabling deformities. I think, therefore, that he was justified in attempting to do something.

We did not have the courage to try this in any other cases in which the thoracic region was affected, but there, obviously, is the place where the deformity is greatest and needs most to be treated. I don't know why there has been such great mystery about the development of the kyphosis. After all, that is what always happens when damage is done to a vertebral body, especially in the thoracic region,—the spine collapses and the kyphosis develops. Our experience has been that you cannot prevent that in these cases by doing a fusion.

In cases of extensive thoracotomy for tuberculosis, frequently a lateral curvature develops with a convexity toward the side of the operation, apparently due to the weakening of the muscles on that side; therefore, extensive removal of the ribs on the side of the convexity would tend to make a curve increase or develop on that side. That is something which must be thought about in making the approach in the thoracic region.

I, too, think that the answer to this problem probably lies in trying to retard growth on the side of the convexity. We have attempted to achieve it gradually by doing a fusion on the convex side of the curve. The

(*Continued on page 170*)

THE EFFECTS OF SHORT-WAVE DIATHERMY ON BONE REPAIR

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Much experimental work has been done on the effects of roentgen ray [9, 18, 19], infra red [20], and ultraviolet [22] irradiation on bone; but there are few references in the literature to the effects of clinical doses of short-wave diathermy on bone repair. Weinberg and Ward noted the increased formation of endosteal bone in holes drilled in canine tibiae for temperature determinations, following irradiation with conventional diathermy. Wise, Castleman, and Watkins noted bone destruction following a single overdose of microwave diathermy in rats. Knudson and Schaible noted that diathermy did not seem to retard bone growth appreciably in white rats. The use of diathermy to stimulate the repair of fractures has been advocated and condemned by many [3, 5, 8, 12, 14, 15, 21, 25], but the actual effects of short-wave diathermy on bone repair have not been studied experimentally.

These experiments were done to determine the effects of short-wave diathermy in clinical or subclinical doses on bone grafts and fracture healing. A standard short-wave diathermy machine was used throughout the investigation.

BONE GRAFTS

Procedure

The dosage of diathermy was standardized to raise the temperature of the medullary cavity of the tibia of a dog weighing thirty-five pounds, 1 degree centigrade after twenty minutes of irradiation. A drum electrode was used, placed at a distance of six centimeters from the leg. The legs were irradiated with a maximum plate current of 150 milliamperes. This standard dosage was used throughout the experiment and was well within the accepted clinical dosage range[3].

In five adult dogs, weighing approximately thirty-five pounds, massive tibial grafts were cut from both forelegs, making a total of ten grafts. The periosteum was raised with the grafts. The grafts were completely severed from their blood supply and were then replaced in their respective beds. Five legs were irradiated twice daily for a period of twenty minutes with the standard dosage of diathermy. The other five legs were used as controls and were shielded with aluminum cylinders to protect them from stray irradiation. One dog was sacrificed at seven days, two dogs at thirteen days, one dog at eighteen days, and one dog at fifty-two days. Cross sections of the entire bone and graft were made.

Observations

It was noted in all sections that new-bone formation arose subperiosteally and gradually bridged the gap between the graft and the bone. In the irradiated legs, the formation of osteoid tissue and new bone was consistently delayed, but it was apparent that the osteoid tissue had calcified as soon as it had formed. The new-bone formation, under the influence of diathermy irradiation, was structurally poor and was laid down without regard for future function.

In the eighteen-day control leg, there was well organized new bone uniting the graft and the bone (Fig. 1-A). This new bone arose chiefly from the endosteum, but a small amount could be seen growing from the periosteum. In the eighteen-day irradiated leg, undifferentiated, dense connective tissue was found between the graft and the bone (Fig. 1-B), and only a small amount of new endosteal bone was seen growing toward the graft site.

After fifty-two days, both grafts were united; but the new bone in the control leg was more mature, and the callus had subsided. The irradiated leg showed considerable callus

FIG. 1-A

Fig. 1-A: Photomicrograph (× 19) of the eighteen-day control leg with the graft on the left. There is marked osteoporosis in both graft and bone and a large amount of endosteal new-bone formation between the graft and bone.

Fig. 1-B: Photomicrograph (× 19) of the eighteen-day irradiated leg with the graft on the right. There is little osteoporosis in either the graft or the bone and undifferentiated, dense connective tissue is seen between the graft and bone.

Fig. 2: Photomicrograph (× 400) of the fifty-two day control graft, showing the revascularization and the formation of new bone within the dilated Haversian canals.

formation and a delay in the healing of the graft. In the irradiated legs, there was also a delay in the reestablishment of the medullary cavity and in the return of myeloid tissue, as contrasted with the control legs. After fifty-two days, the medullary cavity of the control leg had been reestablished, and a considerable amount of myeloid tissue was present. In the leg irradiated for fifty-two days, the myeloid tissue was scanty; and, instead, a considerable amount of endosteal new bone was found.

In all the control legs, there was considerably more osteoporosis than in the irradiated legs. This was shown by weekly roentgenographic examinations and was also shown microscopically by enlargements of the Haversian canals. Osteoporosis did not appear in the irradiated legs until the eighteenth day, and after that time it was minimal. In the control legs, osteoporosis was noticed at the seventh day, and it increased throughout the period of healing.

The osteocytes were present in the lacunae of all grafts for a period of thirteen days. After thirteen days, there were increasing areas in the center of the grafts that showed the lacunae to be empty or to contain pyknotic nuclei. The areas in which these dead osteocytes were found became progressively larger. In the irradiated graft at fifty-two days there were very few osteocytes present, and these were found only peripherally. There

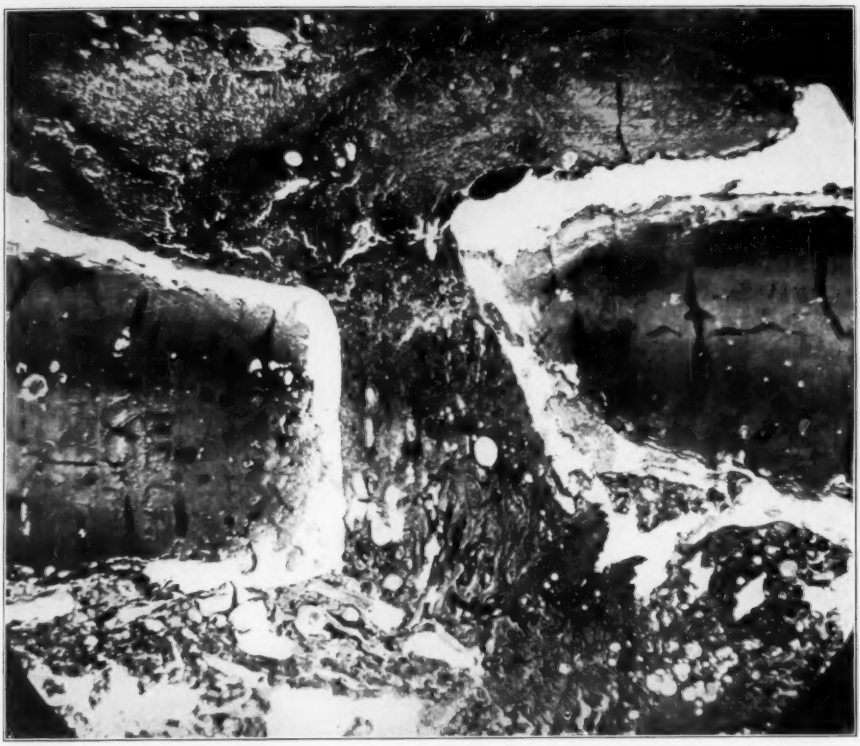

FIG. 1-B

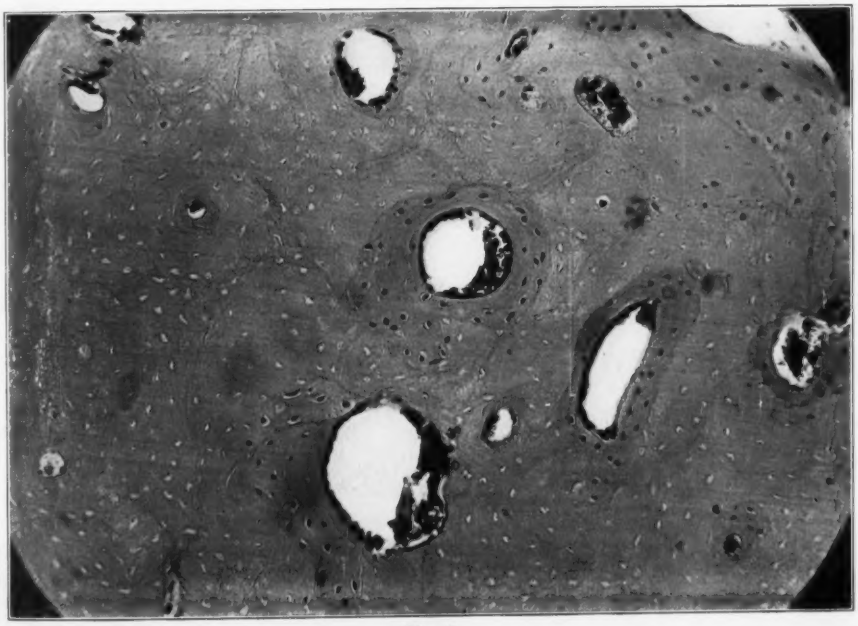

FIG. 2

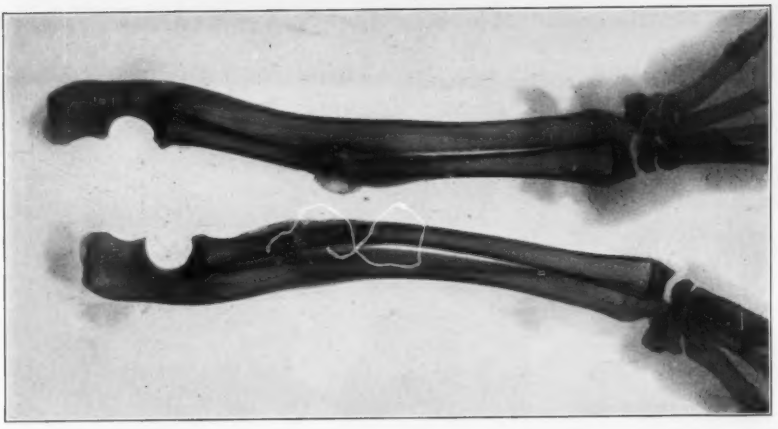

FIG. 4-B

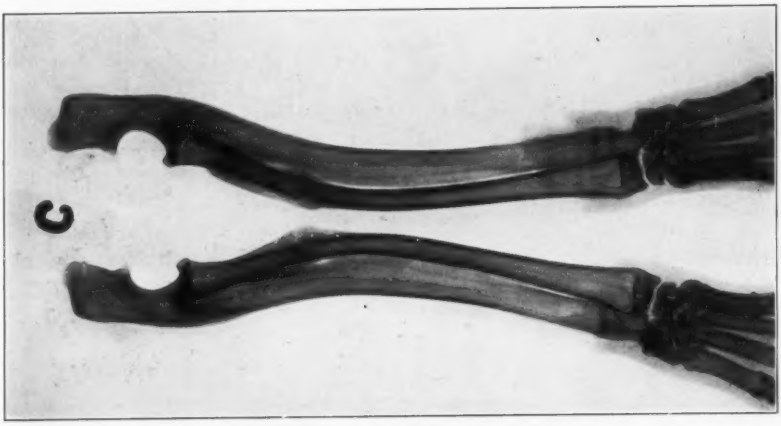

FIG. 4-A

Fig. 4-A: Roentgenogram showing the twenty-one day control fractures.
Fig. 4-B: Roentgenogram showing the twenty-one day irradiated fractures.

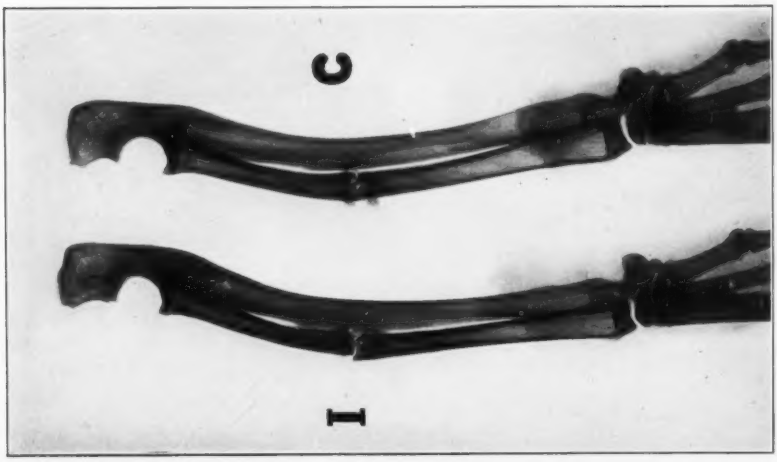

FIG. 3

Fig. 3: Roentgenograms of the eighteen-day irradiated and control fractures, showing increased callus formation about the control fracture site; the irradiated leg is on the left and the control leg is on the right.

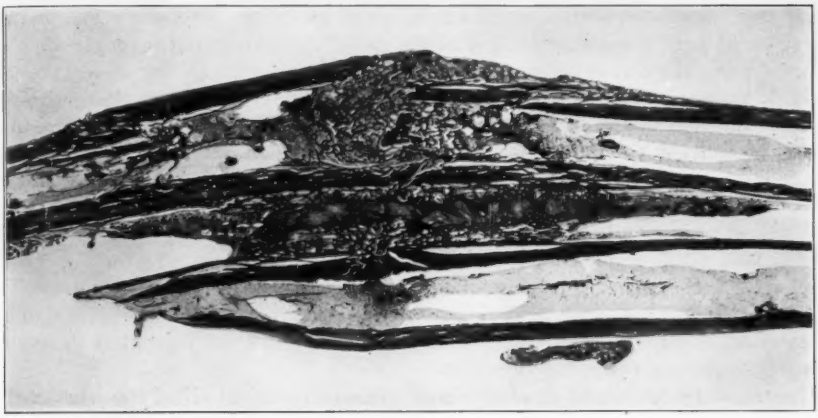

FIG. 5-A

Photomicrograph (× 5) of the twenty-two day control fracture.

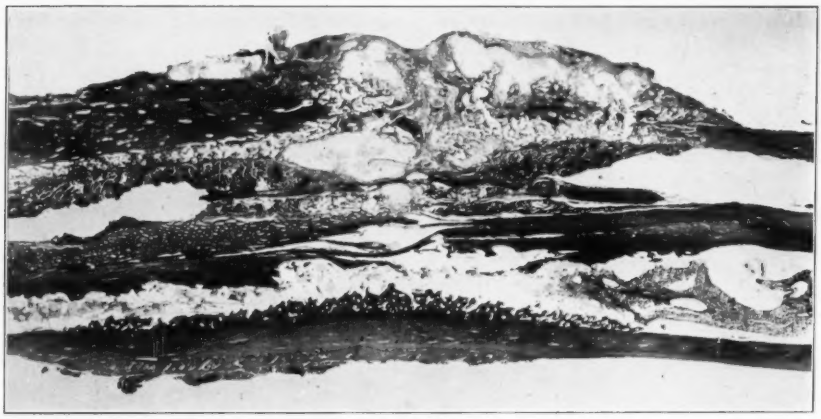

FIG. 5-B

Photomicrograph (× 5) of the twenty-two day irradiated fracture.

were many more osteocytes present in the control graft at fifty-two days but these were found only around enlarged Haversian canals. The dilated Haversian canals seen in the control graft had become revascularized, as seen microscopically by the ingrowth of blood vessels. An abundance of osteoblasts were present, which were forming bone concentrically within the canals (Fig. 2). Few enlarged Haversian canals were seen in the irradiated graft, and the process of replacement of cortical bone was absent.

FRACTURES

Procedure

Twenty-three adult, male rabbits, weighing eight pounds each, were used in this experiment. Both radii of each rabbit were fractured at the midshaft through a small skin incision, a bone-cutting forceps being used. The fractures produced in this manner were transverse and had slightly irregular edges with a minimum of fragmentation. Since the ulna acted as a splint to the radius, no fixation was needed. Five rabbits died during the anaesthesia or were discarded because of fracture of both bones.

Both forelegs of nine rabbits were used as controls. The forelegs of the other nine

rabbits were irradiated with a standard dosage of diathermy for twenty minutes twice daily. The diathermy was discontinued at the end of thirty days. The average rise in skin temperature at the end of twenty minutes of irradiation was 1.8 degrees centigrade as measured with a standard dermalor. The temperature returned to normal within five minutes. One control and one irradiated rabbit were sacrificed at two, five, ten, fifteen, eighteen, twenty-one, twenty-two, thirty, and forty-eight days. Gross observations, microscopic sections, tensile-strength determinations, and phosphatase determinations were made.

Observations

All skin incisions in the irradiated legs showed delay in healing when contrasted with the controls. The two-day and five-day irradiated and control legs showed no significant difference in fracture healing. The hematomata had begun to organize, but there was no evidence of callus or new-bone formation.

Between the tenth and fifteenth days, there appeared in all of the irradiated legs considerable oedema of the skin, subcutaneous tissues, and muscles, which persisted throughout the experiment. The ten-day and fifteen-day irradiated legs showed little callus formation about the fracture sites when contrasted with the control legs. There was very little osteoporosis, and the bones crushed with difficulty. The ten-day and fifteen-day control legs showed no oedema of the skin, subcutaneous tissue, or muscle; but there

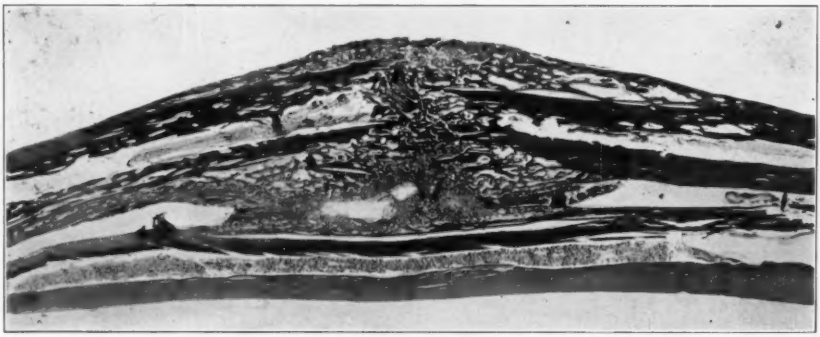

FIG. 6-A

Photomicrograph (× 5) of the thirty-day control fracture.

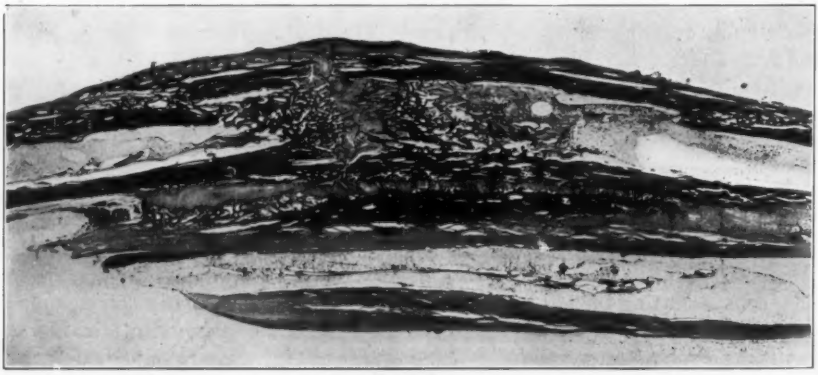

FIG. 6-B

Photomicrograph (× 5) of the thirty-day irradiated fracture.

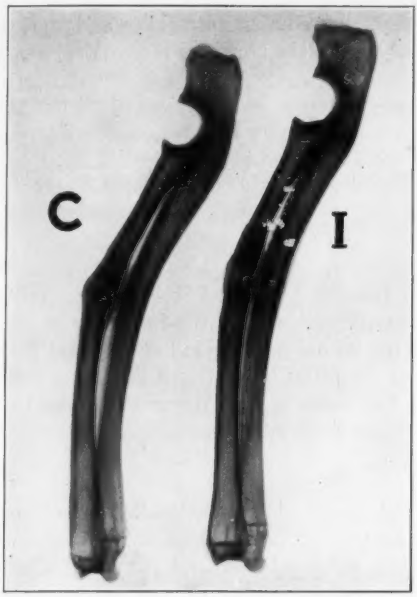

FIG. 6-C

Fig. 6-C: Roentgenogram of thirty-day fractures, showing the control fracture on the left and the irradiated fracture on the right.

Fig. 7: Roentgenograms of the forty-eight day control and irradiated fractures, showing more rapid healing of the control fracture on the right.

FIG. 7

was considerable callus formation extending one centimeter to each side of the fracture site. The bones were osteoporotic and crushed easily.

At eighteen days (Fig. 3), and to a greater degree at twenty-one and twenty-two days (Figs. 4-A, 4-B, 5-A, and 5-B), the irradiated legs showed delayed healing of the fractures and an excess of callus formation. The callus and new bone were of poor quality, were laid down irregularly, and did not show the fusiform shape seen in the control fractures. The control fractures were healing more rapidly and the callus had subsided considerably by twenty-one days. Roentgenograms showed marked osteoporosis in the control legs and a minimal amount of osteoporosis in the irradiated legs. The tensile strengths of these fractures were determined by a pull with weights in the direction of the long axis of the bone. The control fractures broke with twenty-five and thirty pounds, while the irradiated fractures both broke with a weight of ten pounds.

At thirty days (Figs. 6-A, 6-B, and 6-C), the irradiated fractures showed sclerosis about the fracture sites when separated, as contrasted with the fresh bleeding surfaces seen on the separation of the control fractures. New bone had bridged the control fracture, and there was considerable evidence of increased vascularity. The medullary cavity of the bone on each side of the control fracture contained red marrow, while the medullary cavity near the irradiated fracture showed only a semiliquid, fatty marrow. The bone near the control fracture was shown to be osteoporotic by roentgenographic and microscopic examination, but there was little evidence of osteoporosis in the irradiated bones.

At forty-eight days (Figs. 7, 8-A and 8-B), the control fracture had completely healed, the medullary cavity had become reestablished, and the callus had subsided almost com-

pletely. The irradiated fractures at forty-eight days showed abundant callus and marked delay in healing. The control legs were more vascular and the bone was more osteoporotic than in the irradiated legs.

Prior to fracturing the radii, the hair was shaved from both forelegs of all rabbits. After four days of irradiation twice daily for a period of twenty minutes, it was observed that there was much more abundant regrowth of hair on the irradiated legs. This stimulation of hair growth was observed on all irradiated legs throughout the experiment.

PHOSPHATASE DETERMINATIONS

Procedure

Phosphatase determinations were made by removing the bone including the callus and fracture hematoma, extending one centimeter to each side of the fracture site. The bone was carefully weighed, ground, and extracted with a given quantity of water for a period of forty-eight hours, the general technique described by Botterell and King being used. The results expressed in Bodansky units (per gram of bone) were determined by the Lumitron technique with a standard photo-electric colorimeter.

Observations

Two days following the fractures, the phosphatase in both the irradiated and control fractures was less than 0.3 Bodansky unit (per gram of bone) which is the same level that was found in normal bone. After five days, the irradiated and control fractures showed a rapid rise in phosphatase level. There was a slightly greater rise in the phosphatase in the irradiated fractures at five days, but the difference was not considered

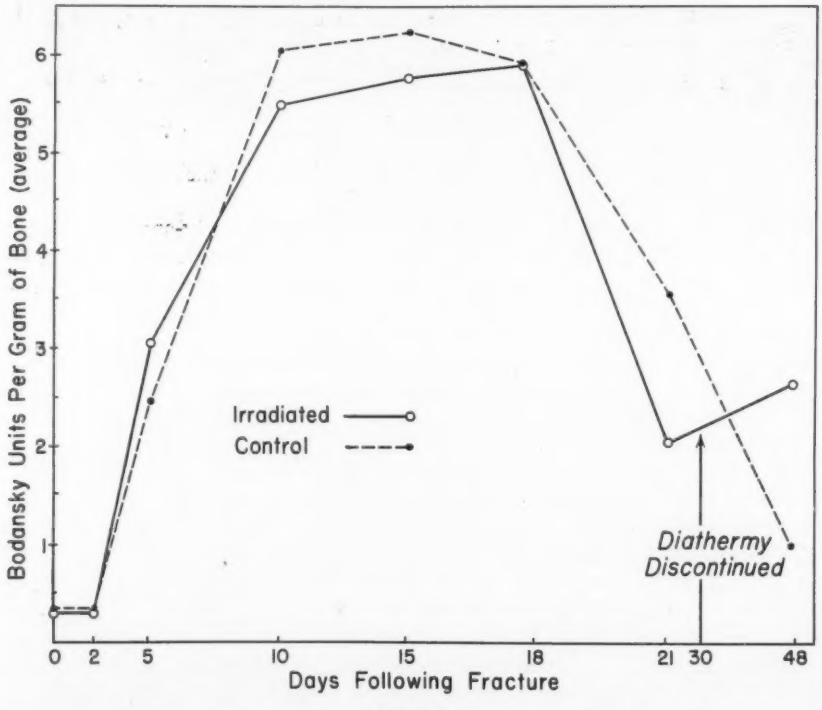

CHART I

Chart showing the phosphatase level of bone at fracture sites.

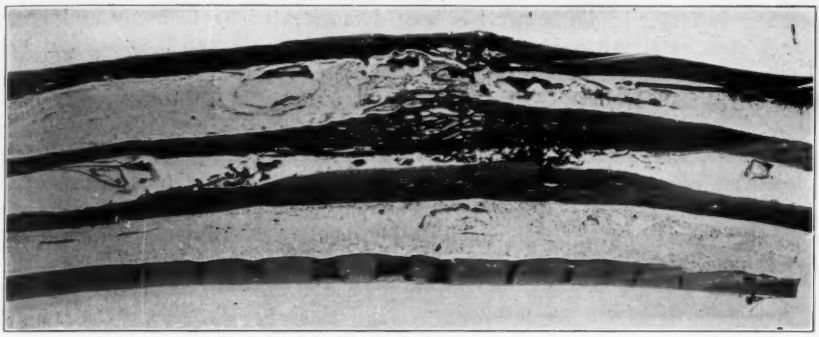

FIG. 8-A

Photomicrograph (× 5) of a forty-eight day control fracture.

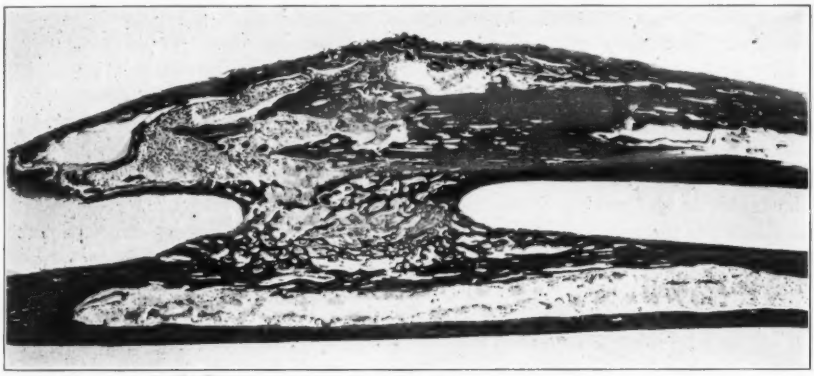

FIG. 8-B

Photomicrograph (× 5) of a forty-eight day irradiated fracture.

significant. Throughout the experiment, the phosphatase levels in both the irradiated and control fractures were strikingly similar (Chart I). Both remained elevated at about six Bodansky units (per gram of bone) for approximately eighteen days, after which the levels began to fall. The phosphatase in the irradiated fractures again became elevated after diathermy was discontinued at thirty days; this rise was compatible with the removal of the inhibiting factor of diathermy in an incompletely healed fracture.

DISCUSSION

It has been shown by these experiments that fracture healing and healing of bone grafts is delayed considerably when diathermy is applied. Under normal stimulation, bone is laid down in an orderly fashion, with the lines of stress developing in accordance with Wolff's law. It was noted in both grafts and fractures that new bone was laid down irregularly which may have been due to modification by diathermy, causing the bone to be laid down without regard to function. There are two factors which may have been responsible: the production of heat and an athermic factor, — possibly, the induction of an electrical current within the bone. Temperature determinations showed that the production of heat was not excessive and was considered to be within the range of safety. Venable and Stuck have shown that an electrical current, due to dissimilar metals near a fracture, will cause delay in the healing of fractures. However, the current induced with diathermy is high-frequency alternating current rather than direct current. The presence

of an athermic factor in diathermy has been demonstrated by Liebesny and Krasny, cited by Kovacs, and has been denied by Knudson and Schaible, and Jacobsen and Hosoi.

Under normal conditions new osteoid tissue is calcified as soon as it is laid down, as shown by Urist and McLean. Although there was a delay in the formation of osteoid tissue under the influence of diathermy, there was no delay in the calcification of the osteoid tissue which had formed. The phosphatase levels per gram of bone at the fracture sites were essentially the same in the irradiated and control fractures. This is in contradistinction to the delay in bone healing under the influence of roentgen irradiation, as shown by Regen and Wilkins, in which the phosphatase activity was greatly inhibited. With diathermy, it appears that the delay in bone repair is due to a delay in the formation of osteoid tissue and not to a delay in the mechanism of calcification of this tissue.

Osteoporosis formation in cortical grafts is shown by the dilatation of the Haversian canals. The dilatation of the Haversian canals in bone grafts is dependent on revascularization, as has been shown by Abbott and his coworkers. It is largely by this method that cortical-bone grafts are absorbed and replaced. The formation of osteoporosis is always associated with increased vascularity, as has been shown by Leriche and Policard, Watson-Jones [11] and many others. It was anticipated that diathermy would cause increased vascularity and possibly stimulate bone repair or cause early formation of osteoporosis. The application of diathermy inhibits the formation of osteoporosis in bone grafts and fractures, and it may be postulated that the delay in the formation of osteoporosis was due to the decreased vascularity which was noted in all irradiated legs.

Since healing is delayed when osteoporosis is minimal, perhaps osteoporosis is an essential factor in the process of normal bone repair. Both bone-graft repair [6] and fracture healing [7] are more rapid in bone showing osteomalacia. Although this is a different process from osteoporosis, as demonstrated by Albright and his coworkers, both show the common factor of a loss of bone substance, producing a more porous cortical bone.

The mechanism of oedema formation following irradiation with diathermy is not known. It has long been known that diathermy increases oedema when present. For this reason, oedema has been listed as a contra-indication to the use of diathermy. The average rise in skin temperature following irradiation with the standard dose of diathermy was only 1.8 degrees centigrade after twenty minutes. This returned to normal within five minutes after irradiation was discontinued. The oedema developed after ten or fifteen days of irradiation twice daily for twenty minutes. Since the temperature gradient decreases from the skin toward the bone with diathermy irradiation, as shown by Osborne and Coulter, the subcutaneous tissues and muscles were heated at less than 1.8 degrees centigrade. For this reason, it is felt that the development of oedema was not due to the production of excessive heat.

CONCLUSIONS

1. Diathermy in repeated clinical doses causes a delay in the healing of fractures in rabbits and in the union of bone grafts in dogs. The new-bone formation and callus are of poor structural quality.

2. Diathermy causes a decrease in the formation of osteoporosis following bone trauma in dogs and rabbits.

3. The continued use of diathermy in clinical doses causes oedema of the skin, subcutaneous tissues, and muscles in rabbits.

4. There is marked stimulation of hair growth in rabbits within four days following the use of diathermy.

5. The use of diathermy causes a marked delay in the healing of surgical wounds in dogs and rabbits.

Note: We are indebted to Dr. T. McD. Anderson, Consultant in Orthopaedic Surgery, and Dr. H. A. Searl, Chief of Physical Medicine, Veterans Administration Hospital, McKinney, Texas, for their valuable assistance in the preparation of this material.

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THE NATURAL COURSE OF OSTEOID OSTEOMA

BY ERIK MOBERG, M.D., GÖTEBORG, SWEDEN

The localized benign disease of the skeletal system, made known by Jaffe as "osteoid osteoma" through a series of articles including a large number of cases, is now almost generally accepted as an entity. It was first described in detail by Bergstrand in 1930. There are now numerous published articles, both of single cases and collective reviews, although comparatively few are from Europe.

Knowledge of several phases of the disease is still incomplete. Its true nature and cause, as well as its natural course, are unknown. The differential diagnosis has likewise been incompletely investigated.

Without doubt a fuller knowledge of its natural course would add insight into the nature of the disease itself. However, the condition is usually interrupted relatively early by operative intervention which, if the nidus has been removed, is always followed by relief from pain and a gradual return to normal of the surrounding eburnated area. Thus the natural course of the disease is not known. Geschickter and Copeland, therefore, on the basis of the morphology alone, could only conclude that one of two things should occur in the typical osteoid osteoma localized in the cortex: "Either the marked periosteal new bone should overwhelm and obliterate the minute rarified lesion, or the active vascular osteoid osteoma should invade the eburnated cortex and produce a progressively larger area of destruction of normal bone. Neither has yet been observed." Geschichter and Copeland were unable to verify the theory that the disease is "a healing area of bone infarction of minute size".

These questions can be clarified by a few histologically verified cases in which the course of the disease has been followed.

CASE 1. A man, aged twenty-three, had had increasing pain for a year, localized in the upper right thigh. Roentgenographic examinations, performed three times during an eight-month period, revealed development of the process, as shown in Figure 1. The diagnosis was verified histologically *.

CASE 2. A woman, aged twenty-four, had complained for over two years of increasing symptoms with

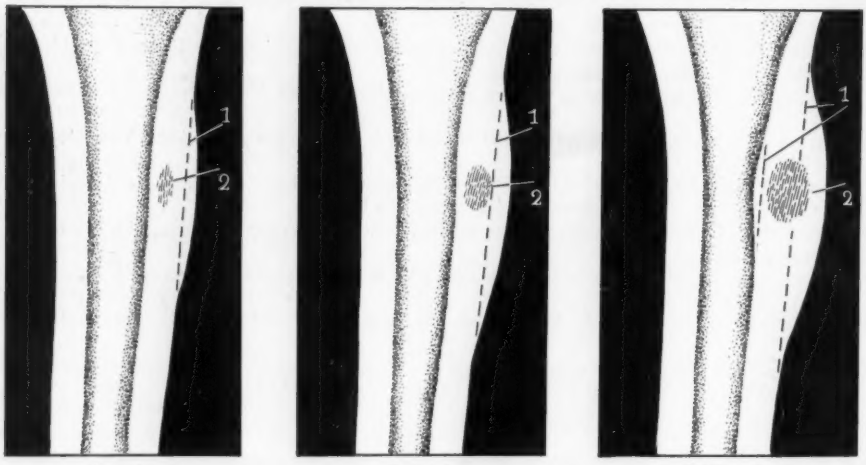

FIG. 1-A FIG. 1-B FIG. 1-C

August 1939 February 1940 April 1940

Case 1. Showing schematically development of the nidus and sclerotic reaction around it.
1: Normal position of cortical border. *2:* Nidus.

* For pictures of histological slide and roentgenograms, see Case 6 in author's previous publication [7].

FIG. 2-A

FIG. 2-B

FIG. 2-C

Case 2. The central rarified area grew in sixteen months, through destruction of surrounding bone, to a size larger in cross section than the normal metatarsal bone at the same place.

Figs. 2-A and 2-B: January 1945.

Fig. 2-C: June 1946.

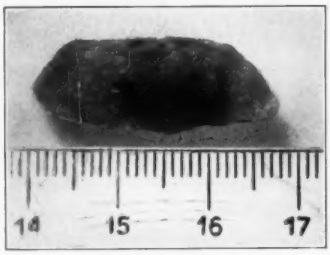

FIG. 2-D

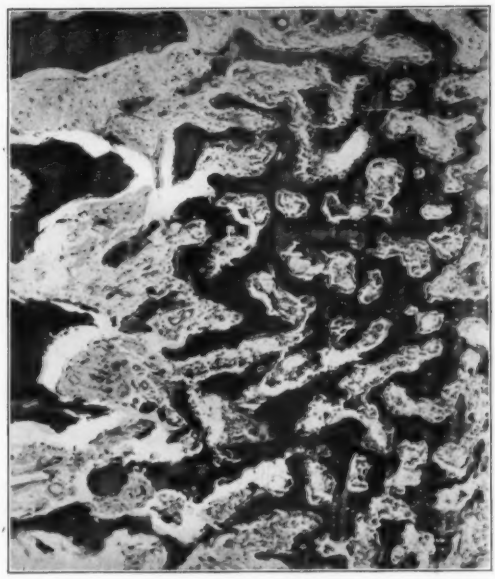

FIG. 2-E

Fig. 2-D: Nidus removed surgically.

Fig. 2-E: Photomicrograph showing tissue at the border between the nidus and surrounding bone.

swelling, pain on walking, and later night pain. The symptoms increased during pregnancy; delivery occurred three months before operation. Two roentgenographic examinations, made sixteen months apart (Figs. 2-A, 2-B, and 2-C) show the development of the process. The sedimentation rate, Wassermann reaction, and blood counts were normal. At operation the findings were typical of the disease (Fig. 2-D), and the diagnosis was verified histologically * (Fig. 2-E). Bacteriological cultures incubated on several media showed no growth.

Mackenzie demonstrated one case in which the nidus in the tibia increased markedly in size in the course of seven months. Sherman also reported one case with the same type of change in the humerus near the elbow, which increased in size during a like period.

All these cases, especially the author's Cases 1 and 2 and Mackenzie's case, show unquestionably that the central rarified lesion developed from a small beginning. *It*

* By C. G. Ahlström.

develops continuously by invasion of normal or eburnated bone. In this manner, as in Case 2, a central area of destruction, larger in cross section than the normal skeleton at the same

FIG. 3-A

Roentgenograms taken in January 1937, showing fifth metacarpal bone one month after the beginning of symptoms. (Case 8 in author's earlier report. Reproduced from *Arch. f. Klin. Chir.*, **202**: 560, 1941.)

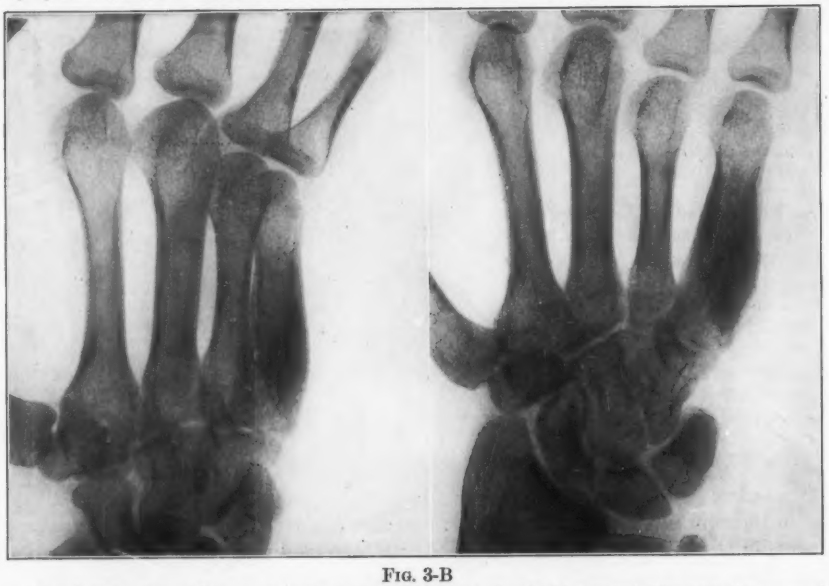

FIG. 3-B

March 1938. Pain still present, but not severe. Bone reaction had increased. The pain disappeared spontaneously a few months later, and check-up roentgenograms in 1941 showed no changes since 1938. (Right view reproduced from *Arch. f. Klin. Chir.*, **202**: 560, 1941.)

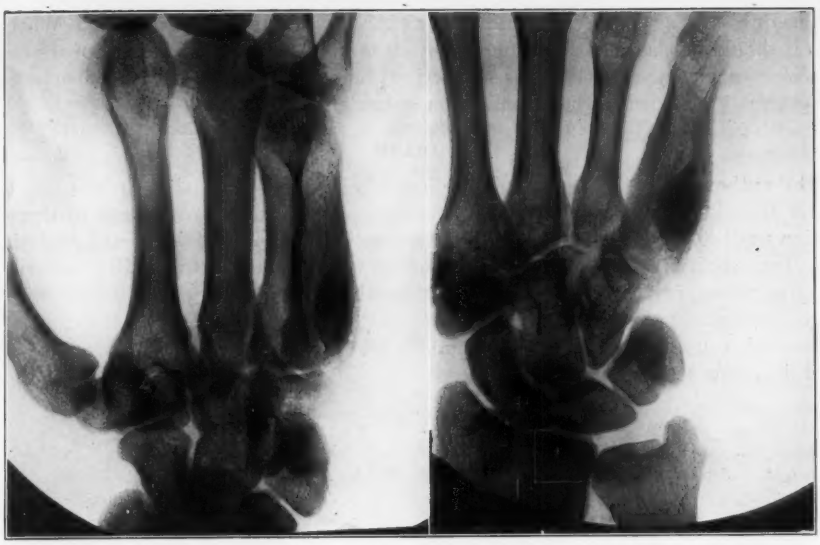

FIG. 3-C

December 1949. Bone reaction slightly diminished. Since 1938 the patient has been entirely free from symptoms in the hand.

place, can develop. This development takes place simultaneously with a progression in clinical symptoms, particularly the night pain.

In view of the fact that, in only eight of 155 cases collected, were the patients over thirty years of age, Sherman concluded that the disease can heal spontaneously. One case published by the author [7] in which the disease was localized in a metacarpal bone, and one reported by Sherman in the tibia, agree both in clinical symptoms and in roentgenographic findings with the typical picture of this disease. The author's patient became free from symptoms without treatment after two years. Check-up examinations were performed four and thirteen years after the first appearance of symptoms (Figs. 3-A, 3-B, and 3-C). Sherman's patient became symptom-free after seven years; the roentgenograms accompanying her article show that the radiolucent area was filled in and had disappeared.

A histological diagnosis is lacking in both of these cases. Unfortunately, it is hardly possible in one and the same case to obtain histological verification of the process and to allow the disease to follow its natural course. A biopsy cannot with certainty be considered to have no effect upon the natural course. Certainly we should consider if these patients had not perhaps been victims of another disease. Certain facts concerning the differential diagnosis [7] show that the roentgenograms in this disease are not wholly typical, and that processes of an entirely different nature could give an almost identical picture. This also explains why some authors do not recognize this condition as a disease entity [2].

However, the diagnosis in the aforementioned cases appears to be so strongly supported by all the facts that they may be assumed to show the natural healing process of the disease. The special remodeling process in the nidus, brought about by the disease, thus *ceases spontaneously as the osteoid bone changes to more normal bone with resulting gradual reduction of the reacting cortical bone mass.* The clinical symptoms presumably disappear with the cessation of progressive development of the central area.

This course, the first half of which may be considered proved, would thus seem to exclude the possibility that the disease is the healing process of a bone infarction.

The questions as to the cause and nature of the disease remain unanswered. Whether or not it should be classified as a tumor depends entirely upon the definition of the term. The picture and course of the disease described herein deviate in several respects from those generally included in the classification of benign neoplasm.

SUMMARY

The author shows, by means of histologically verified cases, that, in the development of the disease process, the small central nidus may increase through further destruction until it may be larger than the cross section of the normal skeleton at a given point. Two clinically and roentgenographically typical, but histologically unverified, cases give presumptive grounds for concluding that the natural course of the disease leads to spontaneous healing. This development appears contradictory to the claim that the disease is caused by bone infarction, and is hardly compatible with the generally accepted definition of a tumor.

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DISCUSSION

Resection of Dorsal Vertebrae in Congenital Scoliosis

(*Continued from page 154*)

difficulty is that only the posterior arches are affected and not the bodies; perhaps if we can find some feasible way of retarding growth in the latter, such as Dr. Nachlas's stapling, that may be the answer. In the case of a single hemivertebra the deformity is not usually very great and, therefore, I think that such a drastic operation as removal is not warranted. In Mr. Wiles's cases, there were multiple hemivertebrae, and those are the ones which result in bad deformities and need to be operated upon.

Dr. John Royal Moore, Philadelphia, Pennsylvania: Mr. Wiles has carefully related his problems with the two patients in whom he excised hemivertebrae. His principal difficulty would seem to be the development of a kyphosis, and he has asked for possible explanations for this development. It would seem to me that there are two principal causes: First, he has destroyed the inferior and superior vertebral plates, which has reduced the growth of the remaining vertebrae to approximately one-half. This would inevitably lead to a loss of body height and to a kyphosis. The second factor, which is perhaps even more important, is that the immobilization was not adequate at the time of his operation and it was not held for a sufficiently long period. Some type of internal fixation should have been employed at the time of the operation and the back should have been held for a minimum of two years.

CONGENITAL DISLOCATION OF THE HIP

Part II. Results of Open Reduction as Seen in Early Adult Period *

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In 1935, Farrell and Howorth described a method of open reduction for congenital dislocation of the hip which had been used successfully in 77 per cent. of 122 hips. The later results in the early adult period of fifty-eight of the hips included in their report are presented here; they represent 47.5 per cent. of the 122 hips. The average follow-up period was eighteen years. The results were evaluated according to the method described in Part I [2]. No other series of adult results of open operation for congenital dislocation of the hip has been found in the literature.

FIG. 1-A

FIG. 1-B

FIG. 1-C

FIG. 1-D

FIG. 1-E

Fig. 1-A: At age of two years, roof obliquity was 35 degrees; well formed false acetabulum.

Fig. 1-B: At age of four and one-half years, roof obliquity was 30 degrees, CE angle 13 degrees. Moderate subluxation and mild coxa valga are shown.

Fig. 1-C: At age of six years, roof obliquity was 15 degrees, CE angle 19 degrees,—a rapid decrease in obliquity and improvement in CE angle.

Fig. 1-D: At age of twelve years, roof obliquity was 15 degrees, CE angle 22 degrees.

Fig. 1-E: At age of eighteen years, roof obliquity was 17 degrees, CE angle 20 degrees. Moderate coxa valga remained; there was persistent mild subluxation.

Complete dislocation and false acetabulum above a slightly oblique acetabular roof (hypoplasia). An adequate acetabular roof developed, despite slight post-reduction subluxation. There was rapid reduction in acetabular-roof obliquity, but acetabulum remained shallow. At age of six years a good result could be predicted. This improvement in the presence of subluxation was seen infrequently (15 per cent.) and occurred only in young patients whose original obliquity was slight.

Adult result: symptomatic 100, physical 90, roentgenographic 94.

* Read at the Annual Meeting of The American Orthopaedic Association, Colorado Springs, Colorado, May 18, 1949. This paper was scheduled for publication in the October 1950 issue, but revision of the manuscript and the illustrations did not reach the *Journal* office in time to make this possible.

TABLE I

STATUS OF REDUCTION IN EARLY ADULT PERIOD

	No. of Hips	Normal	Dysplastic without Subluxation	Subluxation	Dislocation	Reconstruction
Single	30	7	1	19	2	1
Bilateral	28	5	3	18	0	2
Total	58	12 (20.7%)	4 (6.9%)	37 (63.8%)	2 (3.4%)	3 (5.2%)

DEFINITION OF TERMS

Hypoplasia of the acetabulum is applied to that small group of acetabula which have developed normally, maintaining a normal femoral head-acetabular relationship, except that the superior acetabular rim is deficient; this results in a shallow acetabulum and a decrease in the CE angle (Figs. 1-E and 3-H).

TABLE II

COMPARISON OF EARLY ADULT RATINGS IN FORTY-FOUR HIPS WHICH COULD BE GRADED ROENTGENOGRAPHICALLY

	Percentage of Normal					
Rating	100–90	89–80	79–70	69–60	59–50	49–0
Symptomatic	19	10	5	3	4	3
	66		18		16	
Physical	4	13	10	5	5	7
	39		34		27	
Roentgenographic	7	4	9	4	3	17
	25		29		46	

Dysplasia denotes a femoral head and acetabular surface of abnormal conformity with surfaces not parallel, from whatever cause, not necessarily congenital. Subluxation may or may not be present (Figs. 2-E, 2-F, 3-D, and 3-E).

Subluxation denotes any abnormal widening of the medial joint space, with or without upward displacement (Figs. 1-B, 3-F, and 3-H). Shenton's line, or the curve formed by the inferior margin of the femoral neck and head, continuing along the inferior margin

TABLE III

INCIDENCE OF SYMPTOMATIC COMPLAINTS IN FORTY-FIVE HIPS

	Symptoms				
Complaint	None	Intermittent	Mild	Moderate	Severe
Pain in hip	28 (62%)	8 (18%)	7 (16%)	2 (4%)	0
Fatigue	16 (36%)	14 (31%)	6 (13%)	6 (13%)	3 (7%)
Limited activity	18 (40%)	14 (31%)	6 (13%)	7 (16%)	0

of the superior ramus of the pubis, is not considered an entirely reliable guide to the presence of subluxation of the hip. The line is broken as a result of obvious subluxation of the hip (Figs. 2-A and 4-A), but it may not be visibly affected by a minor lateral displacement of the head without upward displacement (Fig. 5-C), and is somewhat affected by the degree of abduction or adduction of the femur. Marked flattening of the femoral head, or a combination of coxa valga and anteversion in the same hip, may result in an interruption of Shenton's line without actual subluxation.

Dislocation denotes a femoral head which is displaced laterally and upward, and receives no visible support from the superior portion of the acetabulum.

FIG. 2-A FIG. 2-B FIG. 2-C

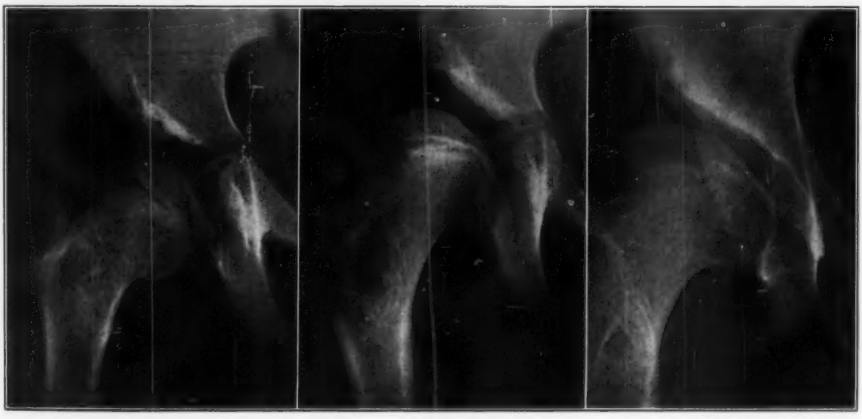

FIG. 2-D FIG. 2-E FIG. 2-F

Fig. 2-A: At age of four years, dislocation and bilabial acetabular roof are shown, with marked anteversion.

Fig. 2-B: At age of four years, with hip in internal rotation, correcting anteversion.

Fig. 2-C: At age of four and one-half years, showing good reduction.

Fig. 2-D: At age of five years, there was slight subluxation.

Fig. 2-E: At age of six and one-half years, acetabular roof was oblique, head flattened; marked coxa valga was present.

Fig. 2-F: At age of twelve years, head was broad and flattened; acetabular support was fair; there was moderate osteo-arthritis.

Complete dislocation with bilabial acetabular roof (dysplasia). A good reduction was followed by subluxation and the development of a poor acetabular roof. A poor adult result was assured at age of six years. An early shelf operation might have prevented this outcome.

Result: symptomatic 100, physical 90, roentgenographic 52.

FINDINGS

The status of reduction of the fifty-eight hips in the early adult period is shown in Table I. Thirty-seven (63.8 per cent.) are listed as being subluxated, but nineteen had minimal subluxation with only widening of the medial joint space, and would ordinarily be included in the normal group (Figs. 1-E, 2-F, and 4-C).

FIG. 3-A FIG. 3-B

A comparison of the ratings in the early adult period (Table II) shows a wide variation between the symptomatic, physical, and roentgenographic results and illustrates the futility of combining these ratings into a common denominator. Sixty-six per cent. of the hips had a grade above 80 per cent. of normal symptomatically, 39 per cent. physically, and 25 per cent. roentgenographically.

FIG. 3-C FIG. 3-D FIG. 3-E

Fig. 3-A: At age of two years, showing dislocation with bilabial acetabular roof.
Fig. 3-B: At three and one-half years, reduction was incomplete.
Figs. 3-C and 3-D: At ages of six and eight years, there was persistent subluxation; roof not developing.
Fig. 3-E: At age of twenty-five years, acetabulum was elongated upward and quite shallow. Only one third of head was engaged. Moderate osteo-arthritis was present.
Complete dislocation with bilabial acetabular roof and non-conformity (dysplasia). The reduction was incomplete, although early; no acetabular roof developed because of the continued pressure of the head against the acetabular margin. Acetabulum was elongated and shallow. At age of four years, a poor result could be predicted.
Adult result: symptomatic 60, physical 60, roentgenographic 30.

At the other end of the scale, 16 per cent. of the hips had a grade below 60 per cent. symptomatically, 27 per cent. physically, and 46 per cent. roentgenographically. A hip with a roentgenographic grade below 60 per cent. of normal can be expected to pursue a progressively downhill course (Figs. 6-A to 6-D), although the rapidity of deterioration varies somewhat with the type and amount of activity, obesity, number of pregnancies, and other factors causing increased weight-bearing or other strain on the hip.

The symptomatic and physical results in the early adult period are shown in Tables III and IV. Sixty-two per cent. of the patients were free of pain and 36 per cent. could

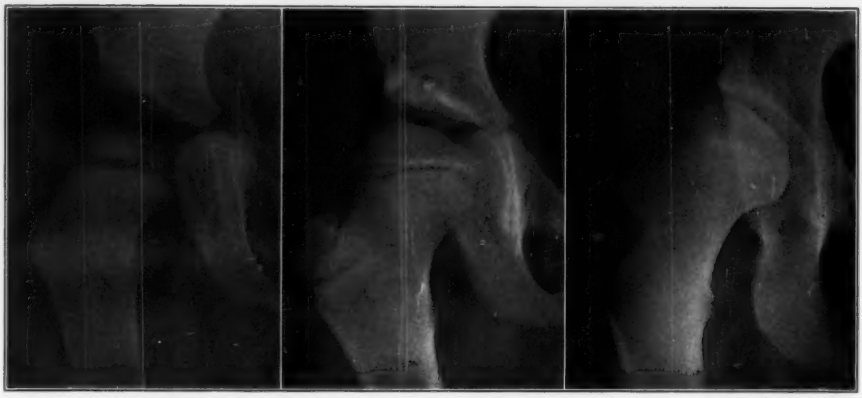

FIG. 3-F FIG. 3-G FIG. 3-H

Fig. 3-F: At age of two years, showing slight subluxation with oblique acetabular roof.
Fig. 3-G: At age of five years, there was persistent subluxation, untreated.
Fig. 3-H: At age of twenty-five years, roof obliquity was 30 degrees, CE angle 10 degrees. Hypoplasia of contralateral hip and slight subluxation (untreated) resulted in a shallow acetabulum with a moderately oblique roof in early adult life. There were no symptoms.
Result: symptomatic 100, physical 83, roentgenographic 75.

not differentiate between the involved joint and the normal joint. Physically, 30 per cent. had a range of hip motion above 80 per cent. of normal, 49 per cent. had no limp, and 86 per cent. had a negative Trendelenburg sign.

TABLE IV

INCIDENCE OF POSITIVE PHYSICAL FINDINGS IN FORTY-THREE HIPS

	None	Intermittent	Mild	Moderate	Severe
Limp	21 (49%)	6 (14%)	12 (28%)	4 (9%)	0
	None	0–½ inch	½–1 inch	1–2 inches	Over 2 inches
Limb shortening	22 (51%)	4 (9%)	10 (23%)	7 (16%)	0
Thigh atrophy	17 (40%)	1 (2%)	12 (28%)	9 (21%)	4 (9%)
	100–90	89–80	79–70	69–60	Below 60
Hip index	3 (7%)	10 (23%)	13 (30%)	5 (12%)	12 (28%)
Trendelenburg	Negative 37 (86%)			Positive 6 (14%)	

Prognostic Value of Periodic Roentgenographic Ratings

A comparison of the roentgenographic ratings made two years after reduction and at puberty (Table V) with the early adult rating reveals that 66 per cent. of the hips had attained their highest grade two years after reduction; by puberty, 85 per cent. had attained their highest grade (Figs. 1-D and 1-E). Improvement after the age of puberty was shown by only 15 per cent. (Figs. 7-E and 7-F).

In adult life 73 per cent. showed a gradual decrease in the grade. A late shelf proce-

TABLE V

TRENDS IN ROENTGENOGRAPHIC RATINGS

	Two Years after Reduction			At Puberty (11 to 14 years)		
	(*No.*)	(*Per cent.*)		(*No.*)	(*Per cent.*)	
Less than adult roentgenographic rating	15	33.3		5	15.1	
Equal to adult roentgenographic rating	5	11.1	66.7	4	12.1	84.9
Greater than adult roentgenographic rating	25	55.6		24	72.8	
Total number of hips graded	45			33		

dure sometimes reverses the downward trend. Hips in which vascular changes developed or which subsequently had reconstructive procedures cannot properly be included in this tabulation.

FIG. 4-A

At age of three years, showing complete dislocation with bilabial acetabular roof.

FIG. 4-B

At age of five years, there was moderate subluxation; roof oblique.

FIG. 4-C

At age of nine years, roof was much better on left, subluxation less; moderate subluxation on right.

FIG. 4-D

At age of nineteen years, roof was good on left, fair on right; very slight subluxation on left, moderate on right.

Complete dislocation and a false acetabulum above a slightly oblique acetabular roof (hypoplasia). An adequate acetabular roof developed, despite slight subluxation after reduction. At age of five and one-half years a poor result seemed assured, yet the acetabular response was much better at nine and one-half years. This result occurred in only 10 per cent. of the hips. A shelf stabilization would seem justified at age of five and one-half years, since 90 per cent. of such hips would otherwise show progressive deformity.

Result: symptomatic 95, physical 80, roentgenographic 75.

FIG. 4-E

The hip passes a crucial period between the ages of five and eight years which is of great prognostic value. Close scrutiny of annual films during this period may reveal a

TABLE VI

EFFECT OF EARLY POST-REDUCTION SUBLUXATION

Condition	No. of Hips	Per cent. of Total	Adult Roentgenographic Rating (*Per cent. of Normal*)
No subluxation	12	21	87
Subluxation	14	25	66
No subluxation, but vascular changes present	5	9	73
Subluxation with vascular changes	20	36	49
Subluxation with shelf reconstruction	5	9	53
Total	56	100	

rapid diminution of an early post-reduction subluxation and obliquity of the acetabular roof (Figs. 1-B, 1-C, and 1-D), or there may be gradual increase in the subluxation and failure of the superior portion of the roof to decrease in obliquity (Figs. 2-D, 2-E, and 2-F). The findings in the hips studied indicate that a change in either direction during this period points to the eventual outcome and is of sufficient reliability to dictate treatment. Marked subluxation of a few hips occurred before this period. Definitive treatment need not be postponed in such hips (Figs. 3-B, 3-C, and 3-D). A few with subluxation showed no change during this period, and must be followed for longer periods, even to puberty, to justify a prognosis. However, the great majority (85 per cent.) in the period from five to eight years indicated a definite trend toward the eventual result (Figs. 1-B to 1-D, 2-C to 2-E, 3-B to 3-D, and 7-B to 7-E).

Effect of Early Post-Reduction Subluxation

Twelve hips which did not become subluxated after reduction had an average roentgenographic rating of 87 per cent. of normal (Figs. 8-A to 8-E). Fourteen with subluxation had an average rating of only 66 per cent. of normal (Figs. 2-A to 2-F and 4-A to 4-D).

TABLE VII

CORRELATION OF POST-REDUCTION SUBLUXATION WITH ACETABULAR-ROOF OBLIQUITY

Status of Reduction	Adult Acetabular-Roof Obliquity	
	Normal	Greater than Normal
No subluxation	12	0
Early post-reduction subluxation	5 (15%)	29 (85%)
Early subluxation with spontaneous recovery	2	0
Late subluxation (after puberty)	1	0

TABLE VIII

INCIDENCE AND RESULTS OF VASCULAR CHANGES

	Vascular Changes			Total
	Mild	Moderate	Severe	
Number of hips	8	14	2	24
Percentage of total hips studied	15.5	24.1	3.4	41
Average number of years followed	13.7	17	18	
Duration of acute changes (years)	2.8	4.4	4.5	
Average adult roentgenographic rating	49	40.4	37	

FIG. 5-A FIG. 5-B FIG. 5-C

Fig. 5-A: At age of seven years, roof obliquity was 55 degrees; roof notched. Anteversion 80 degrees.

Fig. 5-B: At age of seven and one-half years, six months after reduction, there was slight subluxation; anteversion corrected.

Fig. 5-C: At age of eight years, subluxation was increasing.

Fig. 5-D: At age of eleven years, roof obliquity was 40 degrees, CE angle 5 degrees; subluxation increasing. Roof obliquity persisted.

Fig. 5-E: At age of twenty-eight years, hip remained subluxated. Roof obliquity was 45 degrees, CE angle 8 degrees; moderate osteo-arthritis.

Dislocation against superior margin of acetabulum; marked roof obliquity. Reduction good, but not maintained because of defective acetabular roof. Roof failed to develop because of advanced ossification at this age. Failure obvious at age of eight years. A shelf would have helped. Subluxation and high roof obliquity persisted, and acetabulum became elongated and shallow. Moderate coxa valga.

Result: symptomatic 50, physical 70, roentgenographic 25.

When vascular changes were superimposed on subluxation, the rating decreased to 49 per cent. of normal (Table VI). These figures indicate that subluxation is either the causative factor or is invariably a sequel of the causative factor. The other important factor to be considered is a *high acetabular-roof obliquity.* Subluxation and the degree of roof obliquity are intimately associated.

FIG. 5-D FIG. 5-E

Correlation of Subluxation with Acetabular-Roof Obliquity (Table VII)

In the twelve hips which did not become subluxated after reduction, normal adult acetabular-roof obliquity developed in *every* case (Figs. 7-A to 7-E, and 8-A to 8-E). Of the thirty-four hips which became subluxated after reduction, 85 per cent. failed to develop normal roof obliquity in adult life. However, normal adult roof obliquities did develop in five with mild post-reduction subluxation (Figs. 1-A to 1-E and 4-A to 4-E). This emphasizes the importance of observation for a period of months during the crucial period.

The production of a normal acetabular support in each hip which did not become resubluxated suggests that subluxation is the primary factor in producing roof obliquity. When the degree of roof obliquity was divided into three grades—mild, moderate, and

TABLE IX

INCIDENCE OF ANTEVERSION AND RESULTS OF OSTEOTOMY

	Age in Years					Total
	2	2–3	3–4	4–5	Over 5	
Degree of anteversion before reduction						
Normal	1	7	0	2	0	10
Moderate		3	6	5	3	17
Severe	1	7	3	4	10	25
Total						52
Number of osteotomies	1	11	7	6	7	32
Degree of anteversion after treatment						
Normal	2	10	5	5	6	28
Moderate		2		2	2	6
Severe		3	1	1	2	7
Retroverted		2	3	3	3	11
Total						52

severe—post-reduction subluxation occurred in 21 per cent. with mild obliquity, in 50 per cent. with moderate obliquity, and in 95 per cent. with severe obliquity. However, in a number of hips with high obliquity but with adequate reduction, normal obliquity developed in adult life (Figs. 1-A to 1-E and 7-A to 7-F).

Dislocations which are reduced in children under the age of five years more readily maintain reduction without subluxation, due to the rapid development of the acetabular roof (Figs. 1-A to 1-E and 8-A to 8-E). However, when the reduction is carried out after the age of five years, this acetabular response is so retarded that, although adequate operative reduction is performed in the presence of high obliquity, there is recurrent subluxation unless this obliquity is corrected by a shelf stabilization (Figs. 2-A to 2-D and 5-A to 5-D). Figures 5-A to 5-E illustrate the hip of a child first seen at seven years of age, in which there was high obliquity which showed no subluxation six months after reduction; however, three years after reduction the subluxation was marked and the roof obliquity was unchanged. This remained constant until adult life. Figures 9-A to 9-E illustrate a hip of identical appearance in a child of the same age (seven years), in which the obliquity

FIG. 6-A FIG. 6-B FIG. 6-C

Fig. 6-A: At age of four years, roof obliquity was 55 degrees; roof notched.
Figs. 6-B and 6-C: At age of five years, before and after reduction.

was reduced by a primary shelf operation at the time of the original reduction. Subluxation did not develop postoperatively and the hip showed a very good result in adult life.

FIG. 6-D FIG. 6-E FIG. 6-F

Fig. 6-D: At age of five and one-half years.

Fig. 6-E: At seven and one-half years of age. Moderate subluxation had occurred, three years after operation. Roof obliquity was 35 degrees, CE angle 15 degrees.

Fig. 6-F: At age of eleven years, there was persistent subluxation. Roof obliquity was 45 degrees, CE angle 25 degrees.

Fig. 6-G: At twenty-five years, roof obliquity was 45 degrees, CE angle −15 degrees.

Dislocation against superior acetabular margin, with marked roof obliquity. Early subluxation after reduction persisted and produced dysplasia of hip which had elongated shallow acetabulum with thick floor.

Adult result: symptomatic 85, physical 87, roentgenographic 55.

Incidence of Vascular Changes

FIG. 6-G

This subject is being reported separately and will not be discussed in detail here. There are three grades of vascular involvement,—mild, moderate, and severe. Those cases classified as mild had only partial involvement of the epiphysis, usually the medial pole; those with moderate involvement showed general epiphyseal changes with scattered points of increased density; those severely involved had complete avascularity of the epiphysis with fragmentation and later regeneration, as commonly seen in coxa plana.

Vascular changes occurred in twenty-four hips, or 41 per cent. of the fifty-eight hips studied (Table VIII). An additional five hips (8.6 per cent.) were listed as questionable, since the shape of the adult head suggested vascular involvement, but serial roentgenograms in the early months of treatment failed to show the characteristic changes. It is possible that films were not taken at intervals frequent enough to demonstrate minimal involvement.

The groups of mild and moderate cases (Figs. 10-A to 10-H) include twenty-two of the twenty-four hips so involved. The average duration of these changes was less in the mild group (2.8 years) than in the other two groups (4.5 years). Most important, however, is the average roentgenographic rating, which was unsatisfactory in all three groups, ranging from 49 per cent. for the mild cases to 37 per cent. of normal for the severely involved. Only four of the twenty-four hips had a rating over 70 per cent. of normal.

The real damage caused by vascular changes may not be apparent until after puberty,

TABLE X

INCIDENCE OF COXA VALGA

	Before Reduction	Two Years After Reduction	At Puberty	In Early Adult Life
Normal	19			
No subluxation		7	4	7
Subluxation		14	5	7
Coxa vara				
No subluxation			1	1
Subluxation		2	1	1
Coxa valga	3			
No subluxation		3	3	2
Subluxation		7	10	15
Total	22	33	24	33

TABLE XI

EFFICACY OF THE SHELF PROCEDURE

Time of Shelf Operation	No. of Hips	Years of Follow-up	Average Roentgenographic Rating (*Per cent.*)
Early			
Adequate	2	18	63
Poor	5	17	41
Late			
Adequate	4	18	65
Poor	3	15	39

TABLE XII

RESULTS OF GOUGING THE ACETABULUM AT THE TIME OF OPEN REDUCTION

	Not Gouged		Gouged		Gouged to Bone	
	No Vascular Changes	With Vascular Changes	No Vascular Changes	With Vascular Changes	No Vascular Changes	With Vascular Changes
Number of hips	11	6	13	5	10	3
Arthritis of femoral head						
None	7	2	10	0	7	1
Mild	1	2	2	3	2	1
Severe	3	2	1	2	1	1
Arthritis of acetabulum						
None	5	2	6	0	3	0
Mild	4	3	6	3	4	2
Severe	2	1	1	2	3	1
Adult roentgenographic rating	65	61	65	41	65	59

when development of the head and ossification of the acetabulum are followed by osteoarthritis.

Anteversion was found in 80 per cent. (forty-two) of the fifty-two hips in this series on which this measurement was determined (Table IX). Osteotomy for correction of the

FIG. 7-A FIG. 7-B FIG. 7-C

Fig. 7-A: At age of five years, roof obliquity was 55 degrees, anteversion 70 degrees; acetabular roof was notched.
Fig. 7-B: At age of six years, after reduction.
Fig. 7-C: At seven years of age there was slight subluxation.

FIG. 7-D FIG. 7-E FIG. 7-F

Fig. 7-D: At age of seven and one-half years, two years after reduction and supracondylar osteotomy for anteversion, roof obliquity was 25 degrees, CE angle 10 degrees. There was slight subluxation; anteversion corrected.
Fig. 7-E: At age of nine years, roof obliquity was 20 degrees, CE angle 15 degrees. Rapid improvement in roof obliquity.
Fig. 7-F: At age of twenty-five years, roof obliquity was 13 degrees, CE angle 40 degrees. Slight osteoarthritis.
Dislocation against superior acetabular margin with roof of high obliquity. Reduction well maintained. An adequate roof developed eventually, although reduction was not done until child was five years of age. Bilateral club-foot prevented much weight-bearing and may explain the unexpected response. A shelf operation was indicated at time of reduction.
Result: symptomatic 70, physical 70, roentgenographic 100.

anteversion was done on thirty-two of the femora (62 per cent.). Slight retroversion in eleven hips resulted; but for functional purposes these may be considered normal. Therefore, in twenty-nine of the hips (55 per cent.) osteotomy corrected the anteversion. Anteversion, which originally was considered insufficient to justify osteotomy in several hips, persisted in an abnormal degree until adult life (Figs. 8-A to 8-E). The effect of even moderate anteversion in causing resubluxation following adequate reduction can easily be demonstrated at open operation after the head has been reduced; the hip with anteversion of more than 45 degrees slips out very easily unless it is maintained in internal

FIG. 8-A FIG. 8-B FIG. 8-C

Fig. 8-A: At age of two years, roof obliquity was 30 degrees, anteversion 30 degrees.
Fig. 8-B: At age of two and one-half years, showing minimal subluxation but oblique roof, six months after reduction. Hip was in internal rotation.
Fig. 8-C: At age of five and one-half years, roof obliquity was 25 degrees, CE angle 19 degrees. Rapid decrease in roof obliquity.
Fig. 8-D: At age of eleven years, roof obliquity was 17 degrees, CE angle 24 degrees.
Fig. 8-E: At age of twenty years, roof obliquity was 25 degrees, CE angle 25 degrees. Persistent anteversion, shallow acetabulum, no subluxation, and slight coxa magna were present.
Complete dislocation with moderate roof obliquity. A good reduction, well maintained, with good acetabular response. Anteversion of 70 degrees persisted to adult life. External rotation causing appearance of coxa valga.
Result: symptomatic 100, physical 100, roentgenographic 90.

FIG. 8-D FIG. 8-E

rotation. In all such cases the anteversion is corrected before plaster fixation is removed.

Coxa Valga (Table X)

Many hips were not included in the tabulation, either because the angle could not be measured on the roentgenograms which were available, or because deformity due to vascular changes often made the measurement too inaccurate to be acceptable. Excepting these, nineteen hips had a normal angle before reduction and only three were in valgus. Two years after reduction, seven hips which had been incompletely reduced were in valgus position, and fourteen others which had been subluxated had a normal angle. However, as subluxation persisted throughout the years, the percentage of subluxated hips with an increase in valgus rose from 33 to 71 (Figs. 5-A to 5-E); and the percentage with a normal angle decreased from 66 to 33. Those hips which remained adequately reduced, however, showed no essential change from the normal inclination.

Efficacy of the Shelf Operation

A review of the fourteen hips upon which the shelf operation was performed (Table XI) shows that only in six was the shelf technically adequate. A shelf cannot be con-

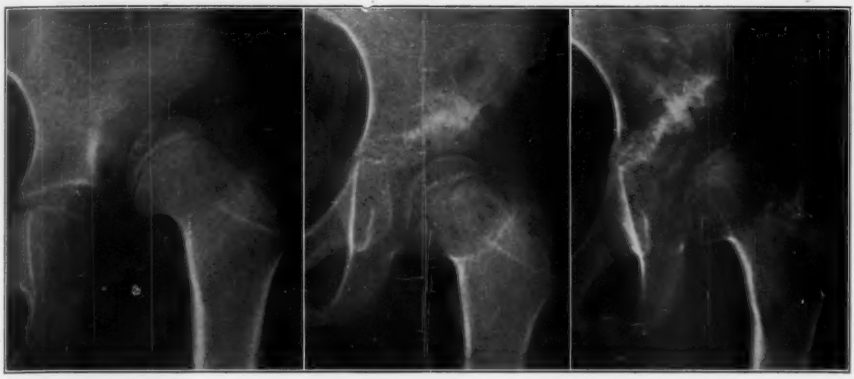

FIG. 9-A FIG. 9-B FIG. 9-C

Fig. 9-A: At age of seven years, roof obliquity was 55 degrees, anteversion 35 degrees. Acetabular roof was deeply notched, floor thick.

Fig. 9-B: At age of nine years, roof obliquity 20 degrees, CE angle 15 degrees. Note effect of shelf applied at time of reduction.

Fig. 9-C: At age of ten years.

Fig. 9-D: At age of fourteen years, roof obliquity was 25 degrees, CE angle 24 degrees. A well formed acetabulum was present.

Fig. 9-E: At age of twenty-six years, roof obliquity was 25 degrees, CE angle 24 degrees. A good result. Slight osteo-arthritis.

Dislocation against the acetabular margin with marked roof obliquity. Open reduction with shelf stabilization carried out; reduction maintained. Compare with Figs. 10-A to 10-I.

Result: symptomatic 100, physical 75, roentgenographic 70.

FIG. 9-D FIG. 9-E

sidered adequate unless it forms a continuation of the acetabular articular surface (Figs. 9-A to 9-E and 10-A to 10-I). A projection some millimeters above the edge of the superior rim cannot effectively support the femoral head unless the head is subluxated. It is apparent that, in all cases in which the shelf was adequate, a satisfactory head-acetabular relationship (normal CE angle) was maintained. All of the early operations were done between the ages of three and six years. It is technically difficult to produce an adequate shelf in a patient younger than five years of age; but, when properly constructed, the shelf remains adequate and materially improves the prognosis.

Effect of Gouging (Table XII)

At the time of open reduction it is frequently found that the acetabulum, even when cleared of the Haversian gland and the ligamentum teres, is too shallow or irregular to retain the head. Cartilage must then be removed from a portion of the roof to form a well rounded socket for the head. This procedure is termed gouging. In thirty-one acetabula (57 per cent.) such treatment was required. Bone was exposed at some point in the floor of the acetabulum in 40 per cent. of these cases. Vascular changes developed

FIG. 10-A FIG. 10-B FIG. 10-C

Fig. 10-A: At age of two years, anteversion was marked.
Fig. 10-B: Also at two years, anteversion was corrected in internal rotation.
Fig. 10-C: At age of two and one-half years, after reduction; slight subluxation was present.

FIG. 10-D FIG. 10-E FIG. 10-F

Fig. 10-D: At age of three and one-half years. Early vascular changes in capital epiphysis.
Fig. 10-E: At age of four and one-half years. Advanced vascular changes.
Fig. 10-F: At age of fourteen years, acetabulum was oblique and elongated. Moderate subluxation.

in 24 per cent. of the group, a figure in proportion to that of the entire series. Osteoarthritis of both the head and acetabulum was no more frequent in these hips than in those which did not have such treatment. Gouging the acetabular cartilage is, therefore, not considered an added risk if it will improve the hip stability. However, in three hips the acetabulum was so shallow that an attempt at excavation was made, the cartilage being removed from the entire acetabulum. This resulted in a stiff hip in each case.

Effect of Age at the Time of Reduction on the Early Adult Result

Table XIII fails to show a clear-cut difference in the symptomatic, physical, and roentgenographic results of hips reduced in children under three years of age as compared with those reduced after the child was three years of age; nor is there any correlation of age at reduction with the incidence of vascular changes. It would seem that age is of less importance when reduction is done by the open method than by closed manipulation, at least up to three years. However, contracture of all structures around the hip increases with age, so that the mechanical difficulty of effecting a reduction is increased, with the added possibility of more trauma and more frequent vascular changes.

FIG. 10-G FIG. 10-H FIG. 10-I

Fig. 10-G: At age of fifteen years. Oblique view of hip showing moderate subluxation.

Fig. 10-H: At age of fifteen and one-half years, three months after shelf operation.

Fig. 10-I: At age of twenty-two years. A good shelf was present. Anteversion persisted.

Dislocation against superior acetabular margin with high roof obliquity. Adequate reduction followed by subluxation and vascular epiphyseal changes at age of four years. A late but adequate shelf improved hip stability. Marked osteo-arthritis resulted from early vascular damage.

Result: symptomatic 95, physical 75, roentgenographic 75 (arthritis).

Incidence of Arthritis

The incidence of arthritis is shown in Table XIV according to the age of the patient at the time of follow-up. Cases in which vascular changes developed are tabulated separately, as the incidence of severe arthritic changes is greater when this complication is present. In the uncomplicated cases, arthritis of the femoral head was fairly evenly distributed throughout the various age groups, indicating that these changes occurred early, before similar changes appeared in the acetabulum. Acetabular arthritis, however, shows a definite increase in degree as the age of the patient increases. The more marked changes are present in the more unstable joints. Obesity in one of these patients makes the specific effect of arthritis difficult to evaluate separately. Nevertheless, a number of patients associated the onset of pain with a gain in weight, particularly during pregnancy.

Grading the severity of arthritis is highly empirical. Minimal changes in both the femoral head and acetabulum were graded as mild (Fig. 9-E). Moderate changes were associated with a decrease in joint space and some irregularity of the opposing surfaces (Figs. 2-E, 2-F, 5-E, and 5-F). Severe changes were associated with gross irregularity in outline or almost complete eradication of the joint space with marked sclerosis (Figs. 3-E and 10-H).

Abnormality of the Contralateral Hip in Cases of Unilateral Dislocation (Table XV)

In seven of the thirty-five hips (20 per cent.), the contralateral joint was hypoplastic or dysplastic when first seen. One of the seven had a normal rating in every respect at early adult age. Two showed symptoms at that time. Four showed definite subluxation, but the joint spaces of the other three were considered normal. In these three, there appeared to be true hypoplasia of the superior acetabular rim. This might be used as an argument in support of the hypothesis that an inherent factor retarding the growth of the superior acetabular rim does exist in this condition. Since six of seven hips retained abnormal features up to early adult life and exhibited factors which predispose to early attritional changes, early and prolonged treatment is certainly indicated.

TABLE XIII

EFFECT OF AGE AT REDUCTION ON ADULT RESULTS *†

Age (*Years*)	Adult Symptomatic Rating	No. of Hips	Adult Physical Rating	No. of Hips	Adult Roentgenographic Rating	No. of Hips
Under 2	75	1	80	1	47	1
From 2 to 3	88	11	80	11	74	15
From 3 to 4	79	4	76	4	49	6
From 4 to 5	87	3	54	3	39	4
From 5 to 6	66	2	73	2	84	3
From 6 to 7		0		0		0
From 7 to 8	50	1	69	1	24	1
From 8 to 9	33	1	40	1	70	1
From 9 to 10 *					50	1

* Not all cases were available for complete examination.
† Cases of avascular change and shelf reconstructions were omitted.

DISCUSSION

In observing the development of these fifty-eight hips from infancy to adult life, some findings were so constant as to be of real prognostic value. Post-reduction subluxation of any degree usually progressed, rarely regressed. In the 15 per cent. of hips that improved after puberty, there was a progressive decrease in the acetabular-roof obliquity in every case. When the subluxation increased, the roof obliquity either remained unchanged at an abnormally high level or gradually increased in degree. In *every* hip which showed no post-reduction subluxation, an essentially normal acetabular roof gradually developed. These changes occurred over a period of five to ten years, but the outcome became obvious before puberty,—usually by the eighth year. Taking into account the effect of anteversion and any vascular change which might have ensued, it was apparent that, after a hip had begun to decline, the adult result would be unsatisfactory unless the trend was changed by altering the acetabular obliquity by a shelf operation.

Although open reduction was done on each hip in this series, it should not be inferred that the authors routinely advocate this in preference to the closed method. Usually the

TABLE XIV

INCIDENCE OF ARTHRITIS IN FIFTY-EIGHT HIPS

	Age at Follow-Up						Total
	14–19 Years		20–25 Years		Over 25 Years		
	Without Vascular Changes	Vascular Changes	Without Vascular Changes	Vascular Changes	Without Vascular Changes	Vascular Changes	
Arthritis of femoral head							
None	5	6	10	5	7	1	34
Mild	2	1	4		2	2	11
Moderate	2	2		1	2	3	10
Severe			2	1			3
Arthritis of acetabulum							
None		1	1				2
Mild	5	2	2	3	4	1	17
Moderate	4	4	7	3	2	2	22
Severe		2	6	1	5	3	17

TABLE XV

VARIATIONS FROM NORMAL OF THE CONTRALATERAL HIP IN SEVEN CASES OF UNILATERAL CONGENITAL DISLOCATION *

	Average Age			
	Five Years (*Degrees*)	Seven Years (*Degrees*)	Twelve Years (*Degrees*)	Adult (*Degrees*)
Average CE angle				
Normal hips	19	20	34	34
Dysplastic hips	10	13	19	11
Average acetabular-roof obliquity				
Normal hips	18	22	10	10
Dysplastic hips	31	24	21	22
Average anatomical rating of the dysplastic hips				81

* These seven hips constitute 20 per cent. of the contralateral hips showing unilateral dislocation.

method of choice is dictated by the age of the patient when first seen. The conclusions of members of this Clinic agree with the majority of reports from other large clinics,—that closed reduction before the age of one year may result in adequate correction in a high percentage of cases. This percentage rapidly decreases after one year of age. However, an attempt at atraumatic closed reduction was usually made in these cases up to the age of three years. If trauma and vascular damage are eliminated, the criteria established for a satisfactory reduction are more important than the manner of reduction. *Any* degree of widening of the medial joint space beyond the limits determined for the normal hip at various ages (Part I, Table VI) is termed subluxation, and any degree of subluxation must be considered serious, especially if it increases gradually.

Anteversion of over 45 degrees enhances the chance for re-subluxation following reduction, and its correction should be considered a routine step in the treatment.

An adequate shelf procedure in the presence of subluxation with an accompanying abnormal acetabular-roof obliquity is effective in reversing the trend toward further subluxation. Its application should be considered as much a routine procedure in the presence of subluxation as is the rotation osteotomy in the presence of anteversion.

Subluxation of the contralateral hip associated with unilateral dislocation should be treated by prolonged abduction therapy. If carried out early and maintained until roentgenograms show acetabular-rim improvement, this will be adequate. Insufficient treatment, or none at all, will result in a symptomatic hip in a rather high percentage of cases in early adult life (28 per cent. in this series). Undoubtedly the percentage rises with each decade of life.

A moderate post-reduction subluxation of a dislocated hip cannot be considered as satisfactory treatment, irrespective of the symptoms and signs of the patient even in early adult life. This study has demonstrated the unreliability of the physical findings as well as the symptoms in detecting the true anatomical status of the hip. A satisfactory functional result for fifteen, twenty, or even thirty years must seem grossly unsatisfactory to a patient who becomes symptomatic in the prime of life. There are many instances of unreduced dislocation (especially bilateral) in which the hips have remained free from symptoms, although admittedly with somewhat restricted activity, for five or six decades. However, examples of moderate to severe subluxation resulting from inadequate treatment in hips which have remained asymptomatic for prolonged periods are much less common. If, then, one is to assume the responsibility for the welfare of the dislocated hip first seen in a child after the age of two years, mindful of a 40 per cent. incidence of vascular involvement with correspondingly poor prognosis, one should not be content with less than an adequate reduction maintained without subluxation.

The objective should be the treatment of these children during the first year of life. Unfortunately, as yet this is not possible in most localities. The results of open reduction in the older group are sufficiently good to justify an open operation for any dislocation which cannot be well reduced or well maintained by closed manipulation. Use of the shelf operation is justified in combination with open reduction when the acetabular roof is defective, or as the primary procedure when reduction without trauma is not possible.

CONCLUSIONS

1. The results eighteen years after open reduction of the fifty-eight hips are as follows:

	Normal (*Per cent.*)	Fair (*Per cent.*)	Unsatisfactory (*Per cent.*)
Symptomatic	65	20	15
Physical	39	34	27
Roentgenographic	17	22	61

2. The optimum roentgenographic grade was attained two years after reduction in 66 per cent. of the hips, and by puberty in 85 per cent. Hips followed roentgenographically in children between the ages of five and eight years show definite trends toward the eventual adult result, a fact which is of practical value in determining further treatment.

3. Vascular changes were seen in 41 per cent. of the femoral heads. In 81 per cent. of the hips so involved, the roentgenographic rating was below 70 per cent. of normal.

4. It is again proposed that an initial capsular relaxation is the primary abnormality in congenital dislocation of the hip. Subluxation and dislocation are the result of the mechanical forces applied to the hip with a relaxed capsule. The acetabular changes are secondary.

5. Any degree of post-reduction subluxation seriously jeopardizes the development of a normal adult hip. Increase of the subluxation ensures an unsatisfactory result. On the other hand, the obtaining and maintaining of perfect reduction ensure an excellent result if no vascular change affects the epiphysis.

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CONGENITAL DISLOCATION OF THE HIP

Part III. Pathogenesis *

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Since Dupuytren's description in 1826, the literature on the pathogenesis of congenital dislocation of the hip has been voluminous, and is replete with confusion, contradiction, and uncertainty. It has recently been sifted in the scholarly work of Badgley. Severin has reviewed the clinical aspects, particularly those presented in the foreign literature. Only

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the bare outline of the principal issues will be presented here, followed by the authors' concept of the pathogenesis, derived from clinical, pathological, and roentgenographic observations. These observations are based on 300 hips in which dislocation was reduced by open operation, fifteen foetal hips dissected between the tenth week and the tenth month of foetal life, and the study of fifty-eight dislocations of the hip reduced by open operation, in which the patients were followed to adult life (reported in Part II).

There is very little knowledge of exactly what happens in congenital dislocation of the hip before birth, and knowledge of the embryological development of the normal hip is incomplete. One hypothesis is that a primary germinal defect in the early stages of embryological development is responsible for congenital dislocation of the hip. However, this idea has been made untenable by embryological studies, such as those of Bardeen and Lewis, and more recently by that of Strayer. In 1943, Strayer described the basic embryological development of the hip and showed that the joint forms as a hemispherical cleft in a block of cartilage, making dislocation impossible before the 35-millimeter stage of the embryo, when the hip joint becomes well defined. Studies [5] of fifteen foetal specimens indicated that the hip joint is well formed by the tenth foetal week, when the acetabulum, femoral head and neck, and capsule are well developed. The normal embryological and foetal development of the hip, therefore, would tend to ensure a stable joint.

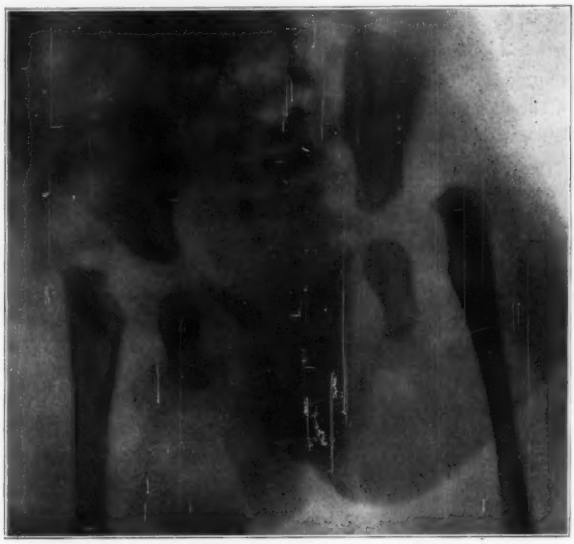

FIG. 1

Bilateral complete dislocation of hip at eight months of foetal life. The acetabular roofs are no more oblique than in normal hips at this age. (Reproduced, by permission of J. B. Lippincott Company, from Ann. Surg., 125: 221, 1947.)

Gross deformity of the hip, associated with congenital club-foot, arthrogryposis, or other congenital deformity, is not considered to be the same abnormality as simple congenital dislocation of the hip. Other developmental factors, such as differences in the male and female pelvis, which might account for the sex incidence of the condition, anteversion, and various mechanical stresses incident to the assumption of the erect posture are considered by some as primary factors; however, most workers, including the authors, consider them secondary. The most widely accepted theory of the cause of the dislocation is that of primary dysplasia of the acetabulum with high roof obliquity, which provides inadequate support for the femoral head.

There are, however, many contradictions to the theory of primary acetabular dysplasia. Roentgenograms of a bilateral complete dislocation of the hip at the eighth foetal month show normal acetabular-roof obliquity for this age (Fig. 1). The authors have seen one unilateral subluxation and one bilateral dislocation in the first week of life, also with normal roof obliquity. These cases further show that dislocation of the hip sometimes occurs before birth.

The only foetal autopsies showing uncomplicated hip dislocation of which we are aware were reported by Ortolani. The principal findings in the three specimens reported

by him were a relaxed capsule and deformity of the posterior cartilaginous acetabular rim, permitting subluxation and dislocation posteriorly. Roentgenograms of the specimens were particularly deceiving; one case, which roentgenographically showed a high acetabular-roof obliquity, anatomically revealed a roof and glenoidal labrum normal in development and providing a good mechanical support to the femoral head after reduction. Another specimen with a roentgenographically normal obliquity had a less well defined labrum and anatomically seemed less stable. Ortolani concluded that all intra-uterine dislocations took place over or through the posterior labrum and were not affected by the stage of development of the superior acetabular roof.

This concept of primary acetabular dysplasia cannot explain the repeated observation in some cases of complete dislocation with the formation of a false acetabulum above a superior acetabular rim which is only slightly oblique (Figs. 1-A, 2-A, 7-A, and 8-A in Part II), while in other cases the acetabular obliquity is much greater and yet the femoral head remains at the level of the defective roof and does not ride above and form a false acetabulum (Figs. 3-A to 3-E, 5-A to 5-E, and 9-A to 9-E of Part II). If there were genetic dysplasia of the acetabular roof, why should every hip in the series reported in Part II in which adequate reduction was obtained and maintained have achieved normal acetabular support in adult life (Figs. 7-A to 7-F and 8-A to 8-E in Part II); and yet 85 per cent. of the cases in which reduction was incomplete or subluxation was permitted to recur have failed to achieve normal acetabular support by the end of the growth period (Figs. 2-A to 2-F, 3-A to 3-F, and 4-A to 4-E in Part II).

The concept adhered to in this report and previously [5] is consistent with all the known pathological and roentgenographic findings in congenital dislocation of the hip. Capsular elongation or relaxation is assumed to be the primary etiological factor. Widening of the joint space medially is not permitted in any normal hip, as demonstrated in dissections of the hip of the normal foetus and confirmed by push-and-pull roentgenograms (Figs. 2-A and 2-B) of young infants. Elongation or relaxation of the capsule is essential to lateral displacement of the femoral head. Lateral displacement is necessary before dislocation can occur. *In utero* the hip is in flexion; with the capsule relaxed, pressure applied along

FIG. 2-A

Unilateral dislocation in an infant of two months, upward pressure being exerted. The head is lateral to the acetabular margin and the acetabular obliquity is only slightly more than in the normal hip.

FIG. 2-B

The same child, with traction. The unossified head has been pulled down opposite the superior acetabular margin. The push-pull films demonstrate the amount of capsular relaxation.

the long axis of the femur would produce a pressure deformity of the glenoidal labrum posteriorly and inferiorly, and the femoral head might be forced out of the acetabulum at this site.

If capsular relaxation and mechanical forces do not effect a definite dislocation *in utero*, one of three courses takes place before birth, depending upon the degree and duration of capsular relaxation:

1. Relaxation may be so *slight* as to permit only subluxation, which, as demonstrated by Putti, may (a) progress as a result of weight-bearing to complete dislocation, (b) persist as subluxation (Figs. 3-F to 3-H in Part II and Fig. 3 in Part III), or (c) spontaneously revert so as to produce a hip of normal appearance.

2. If relaxation is *moderate*, the head may be partly retained within the acetabulum until normal muscle tone and weight-bearing produce sufficient pressure in the long axis of the femur to force the head against the periphery of the plastic superior acetabulum, resulting in the gradual formation of a fossa or in a bilabial appearance of the superior acetabular rim (Figs. 3-A and 7-A in Part II).

3. Finally, if capsular relaxation is *severe*, the head becomes dislocated over the intact glenoidal labrum and is lodged above it, creating a false acetabulum by abnormal pressure against the ilium (Figs. 1-A, 5-A, 6-A, and 9-A in Part II). Sometimes this even forces the labrum downward, as seen at operation in some of the authors' cases and as demonstrated clearly by Wiberg in his arthrographic studies.

While the femoral head is displaced laterally and upward, as seen at operation in numerous cases, the superior portion of the capsule is pushed ahead of it, so

FIG. 3

Congenital subluxation of hip persisting to adult life. The lower portion of the acetabulum has become thickened and has filled in, but its original location can be made out. The acetabular roof is high and somewhat oblique, and the contour of the head and the roof conform to each other. Nearly half of the head is displaced lateral to the acetabulum.

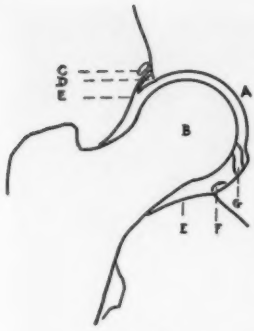

FIG. 4-A

FIG. 4-B

A. Acetabulum. *B*. Femoral head. *C*. Rectus tendon. *D*. Glenoidal labrum. *E*. Capsule. *F*. Transverse ligament. *G*. Ligamentum teres.

Fig. 4-A: Head normally situated in acetabulum, as in infancy or adult life. The capsule is not relaxed, but the ligamentum teres is relaxed except at the extremes of rotation or adduction. The head is secure under a good roof. The C-E angle and acetabular obliquity are normal.

Fig. 4-B: Slight subluxation, with excessive pressure against superior acetabular margin and glenoidal labrum, which is turned up. The capsule is slack and elongated; the ligamentum teres is less slack. The C-E angle and the acetabular obliquity are increased. The head slips back and forth with weight-bearing and changes in position.

FIG. 4-C

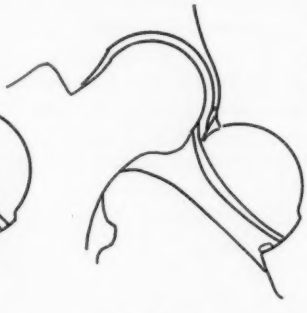

FIG. 4-D

Fig. 4-C: Marked subluxation, with excessive pressure against superior acetabular margin and glenoidal labrum. Much of the weight is carried by the capsule, which is elongated but taut; the ligamentum teres also is taut. The superior acetabular margin and glenoidal labrum are folded back, elongating the acetabulum and increasing its obliquity. The C-E angle is increased. The head is flattened medially and elongated downward to conform to the contour of the elongated acetabulum and the pull of the capsule inferiorly.

Fig. 4-D: Complete dislocation. The head and capsule are pressed against the ilium above the acetabulum; the capsule often becomes adherent to the ilium, is transformed into fibrocartilage, and becomes the "false acetabulum". The capsule and ligamentum teres are elongated and taut. The inferior portion of the capsule is pulled up over the lower acetabulum like a curtain, preventing re-entry of the head.

that, with a complete dislocation, the capsule is finally pressed against the ilium and often becomes adherent to it, participating in the formation of the false acetabulum. The inferior portion of the capsule with the transverse ligament is pulled up like a curtain over the inferior portion of the acetabulum, eventually closing off this portion sufficiently to prevent readmission of the head (Figs. 4-A to 4-D). There is no hourglass constriction, for there is usually no side-to-side constriction and there is never constriction superiorly. The ligamentum teres, which normally acts as a checkrein, limiting adduction and rotation of the hip, also acts as a deterrent to upward dislocation, but normally has enough slack to be ineffective in preventing lateral displacement and subluxation. In dislocations it always becomes elongated and, unless it degenerates and ruptures, may by its bulk interfere with reduction.

The exact time of dislocation has not been demonstrated in any case, but it appears likely that the dislocation is gradual rather than sudden. There are certain known findings which may have some bearing on this point. No dislocation in which the superior roof of the acetabulum presented a bilabial appearance or in which a fossa had formed at the superior roof of the acetabulum has been seen by the authors in infants before the age of nine months, although we have seen about forty subluxated or dislocated hips in infants below this age. This suggests that the duration and degree of pressure against the acetabular roof has been insufficient to produce deformity; that ossification of the acetabular rim is insufficient to show the deformity which is present only in the cartilaginous structures; or that capsular elongation is of such degree as to permit complete dislocation above the superior acetabu-

lar rim. The authors have noticed repeatedly that the acetabular obliquity in the early cases (infants younger than eighteen months) (Figs. 2-A and 2-B in Part III) is not so severe as in hips with a well formed superior acetabular fossa (Figs. 2-A and 7-A in Part II). Also, in those children over eighteen months of age with a well formed false acetabulum, the superior roof obliquity is about the same as the normal roof obliquity of an infant one to six months of age (Figs. 1-A, 5-A, 6-A, and 9-A in Part II and Figs. 2-A and 2-B in Part III).

It is concluded from these observations that, if the femoral head is permitted to dislocate completely in the early months of life, there is no pressure deformity of the acetabular rim and, therefore, the roof obliquity remains essentially as it had been at the time of dislocation (Figs. 1-A to 1-E, 7-A to 7-F, and 8-A to 8-E in Part II). In time the floor of the acetabulum thickens as the result of the absence of the pressure of the femoral head, and this may gradually increase the obliquity by filling from within (Fig. 3). However, it has been repeatedly demonstrated that, if the head can be adequately reduced and maintained at this early age, the acetabular roof develops rapidly to a normal appearance (Figs. 8-A to 8-E in Part II).

It has also been demonstrated that, even in children up to the age of at least five years, the acetabulum retains this ability to respond to the presence of the femoral head in the normal position and to produce an adequate acetabular roof (Figs. 7-A to 7-F in Part II). The older the patient, the longer the time required for the development of an adequate acetabular support. In older patients, resubluxation may occur before acetabular support has developed (Figs. 9-A to 9-E in Part II), unless the normal process is aided by the formation at operation of a shelf to assist in the maintenance of reduction.

In summary, the cartilage of the superior acetabular margin and the glenoidal labrum is quite plastic in the foetus and the infant. As soon as subluxation occurs, the pressure of the femoral head against the glenoidal labrum and the superior margin of the acetabulum depresses them and a groove or fossa begins to form, resulting in the oblique acetabular roof. Unless the pressure is relieved, the obliquity increases, and when ossification occurs, at about the age of five years, the obliquity is permanent. Continued pressure of the head against the superior margin and filling in of the base of the acetabulum cause the acetabulum to become elongated and shallow. Complete dislocation above the acetabular margin, with no pressure against the margin, results in a small acetabulum due to filling in, but obliquity of the roof is relatively slight or absent.

The cartilage of the femoral head of the infant is also plastic, and the shape of the head is determined by its relation to the acetabulum or the ilium. Pressure of the head against the superior margin of the acetabulum results in flattening of the head medially and protrusion inferiorly (Figs. 3-A to 3-F and 5-A to 5-E in Part II). When the head is dislocated against the ilium above the acetabulum (Figs. 1-A to 1-E and 9-A to 9-E in Part II) or posteriorly, it usually moves freely and has little or no bony support, retaining its spherical shape. When it remains firmly against a false acetabulum, however, the shape of the latter and of the head tend to conform to each other (Figs. 5-A to 5-E in Part II). In an older child, a head which was originally spherical tends to remain so even when subluxated against an oblique acetabulum (Figs. 6-A to 6-G in Part II). It is difficult to determine how often and to what extent vascular changes are responsible for the molding effect.

It is of interest to compare the deformities of other conditions of the young hip with those of congenital dislocation. Approximately half of the hips involved by coxa plana show slight subluxation or widening of the medial joint space. This subluxation is permanent in approximately one-quarter of the cases. In the hips with permanent subluxation, a definitely abnormal obliquity of the superior acetabular roof, closely simulating the appearance of treated congenital dislocation in adult life (Fig. 5), develops in some cases. When the gluteal muscles have been paralyzed by poliomyelitis in infancy or early child-

FIG. 5

FIG. 6

FIG. 7

Fig. 5: Coxa plana with subluxation. The acetabular changes are similar to those of congenital subluxation.

Fig. 6: Dislocation following paralysis of hip muscles due to poliomyelitis at an early age. There is considerable deformity of the acetabulum, similar to that seen with congenital dislocation, and flattening of the head medially. The capsule is elongated.

Fig. 7: Subluxation associated with spastic paralysis, with acetabular changes similar to those of congenital subluxation. There is also considerable coxa valga and early osteo-arthritis.

hood, the unsupported capsule is stretched by the force of weight-bearing, and subluxation or even dislocation occurs. In the presence of this subluxating force, the acetabular roof fails to develop normally and, in some cases, is as badly deformed as in severe congenital dislocation (Fig. 6). Similarly, in spastic paralysis there is occasionally such severe adductor spasm that subluxation is produced and acetabular-roof obliquity results. Thus deformities of the acetabulum, similar to those seen in congenital dislocation of the hip and demonstrably due to relaxation of the capsule and pressure against the acetabular roof, are seen in other subluxations of the hip in infancy and early childhood.

A number of constant anatomical changes associated with congenital dislocation of the hip are thought by the authors to be entirely secondary to the prolonged subluxation or dislocation. The most important of these are anteversion, coxa valga, and weakness of the thigh and hip muscles. Vascular changes of the epiphysis might be considered in this category, but they are more commonly the result of treatment, although residual deformity, typically due to vascular changes, has been noted in hips which were never subjected to manipulation or operative reduction. Vascular changes are second in importance only to adequate reduction in the treatment of this condition. A discussion of the cause and effects of these changes is the subject of a separate report, since even the minimal and apparently transitory vascular changes bear ominous forebodings.

Anteversion of the femoral neck in relation to the shaft in the foetus and infant is usually 20 to 40 degrees, more than twice that of the adult. Anteversion in the usual antero-superior dislocation is commonly between 30 and 70 degrees, but retroversion has been found in posterior dislocations. Anteversion is considered to be largely torsion of the shaft, the head and neck remaining in the normal relationship with the trochanters, as is well demonstrated in a bilateral case with unilateral osteotomy (Figs. 8-A to 8-E in Part II). In the infant hip, reduction may be followed by gradual diminution of the anteversion, as observed by Gill and by Howorth; but certain patients in this series with moderate to severe anteversion, not corrected by osteotomy, maintained an abnormal amount of torsion until adult life (Figs. 3-A to 3-F, 8-A to 8-E, and 10-A to 10-I in Part II).

Coxa valga, as noted in Table X (Part II) was present after reduction in some degree in from 25 to 60 per cent. of the cases. The measurement of this angle is subject to con-

siderable technical error, since varying degrees of rotation of the shaft affect greatly the appearance of the angle in the roentgenogram. To obtain an actual measurement there must be a true anteroposterior view of the neck, obtained only after sufficient internal rotation of the lower extremity to counterbalance anteversion (Figs. 3-A and 3-B in Part II). The angle of the neck with the shaft was taken to be the angle formed by a line through the axis of the shaft with a line parallel to the axis of the neck, but passing through the center of the articulating surface of the femoral head.

No close correlation could be seen in these hips between the frequency of post-reduction subluxation and the presence of coxa valga. It was noticed, however, that in those cases in which reduction was carried out before the age of three years and the position was maintained, coxa valga did not develop. Those untreated hips in which subluxation persisted to adult life almost invariably showed moderate to marked degrees of coxa valga (Figs. 2-F to 2-H in Part II). Inman explained the mechanics of the development of this deformity as the normal reaction to stress (Wolff's law) as the hip is abducted and the trunk tilted toward the affected side. This causes the reacting force vector (total of the hip abductor force vectors) more nearly to coincide with the mechanical axis of the lower extremity, thus counteracting the influence of both subluxation and weak abductors.

The muscles of the congenitally dislocated hip are well developed, but the effect of the poor fulcrum provided by the unstable hip is to weaken the effective strength of those muscles which are relaxed by this instability. They tend to assume normal strength and function after a successful reduction, although one of the most constant observations in adults with otherwise normal physical findings was atrophy of the thigh and abductor weakness.

Since the one constant pathological finding in congenital dislocation or subluxation of the hip is elongation or relaxation of the capsule, this is considered to be the primary etiological factor. The cause of the elongated capsule is unknown; it is possibly of genetic origin, possibly mechanical, possibly hormonal, or due to some nutritional deficiency as yet unknown. It is frequently associated with general relaxation of the ligaments of other joints, particularly of the feet. Why is not the incidence of hip dislocation higher in children with severe pes calcaneovalgus? Why is the incidence of congenital dislocation so predominantly female? Why is the condition usually unilateral? Why do some subluxations in infancy spontaneously return to normal, while others get worse? Why does the mere maintenance of correction in abduction for short periods in infancy suffice to permanently interrupt the cycle, terminating in dislocation and pressure deformity of the superior acetabulum? We can answer these questions only with suppositions. However, inability to explain all the facts does not detract from the usefulness of the only theory which does not contradict any of the anatomical findings in congenital dislocation of the hip, and which seems useful in interpreting many of the clinical findings.

Admitting capsular relaxation to be the primary etiological factor, and assuming that all reparative processes of the hip joint are normal and dependent on only one condition—obtaining and maintaining a normal head-acetabular relationship—the problems of this condition seem greatly simplified and almost wholly within our control. If, at the same time, we can prevent the occurrence of vascular changes in the capital epiphysis, or adequately treat these changes once they have been recognized, the results of early treatment of congenital dislocation of the hip should be excellent even by the highest standards.

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THE MEDICAL-CARE PROGRAM OF THE NATIONAL FOUNDATION FOR INFANTILE PARALYSIS, INC.*

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Prior to discussing the medical-care program of The National Foundation for Infantile Paralysis as it is carried out today, it seems well to review briefly the history and development of the National Foundation since its incorporation in 1938. Some of you who are members of The American Academy of Orthopaedic Surgeons shared in the early establishment of the Foundation and its medical program. Of the men and women who participated in this voluntary health movement, I am sure none anticipated that this one agency would, in a single year, spend approximately $31,000,000 in combating an epidemic of poliomyelitis and in financing care for patients who had suffered in previous epidemics.

The National Foundation may be likened to a business enterprise with some 2,800 branch offices, representing chapters of the National Foundation in practically every county of the United States. The sole financial support of the National Foundation comes from the annual "March of Dimes", which is held the last two weeks of January. Of the funds raised by the chapters, 50 per cent. remains in the local area to finance care of patients, either for direct payment for services or the purchase and procurement of equipment for hospitals, so that care for patients may be readily available. Since the charter of the National Foundation does not permit its operation of hospitals or the direct care of patients, my discussion today will be primarily an outline of a local chapter's function and its expenditure of the 50 per cent. of the funds that it retains.

In 1938, there was no established precedent to serve as a guide for The National Foundation for Infantile Paralysis in establishing a medical-care program. There were, of course, purely local programs of a voluntary nature and many official programs under which medical care was made available to the indigent and to the aged. The program of the local chapter has depended largely upon the guidance that has been given to the chapter by the Medical Advisory Committees of the counties. The local chapters are staffed entirely by volunteer workers, which has made it impossible to establish an over-all national program.

*Read at the Annual Meeting of The American Academy of Orthopaedic Surgeons, New York, N. Y., February 14, 1950.

Unlike some voluntary agencies, The National Foundation for Infantile Paralysis has never dictated to physicians regarding the treatment of acute, convalescent, or chronic poliomyelitis. We believe that the attending physician is the one best qualified to prescribe treatment for his patient. Through an educational program and the stimulation of clinical research through grants-in-aid, we have attempted to bring to the medical profession of this country a knowledge of newer developments and methods in treatment, knowing that the physicians themselves would select that which is good and discard that which is bad.

The National Foundation chapters have but one obligation,—to see that no patient is denied the medical care ordered by his attending physician because of inability to meet the cost. The funds of the National Foundation are, in fact, public funds, even though they are raised by voluntary contribution as opposed to the health programs which are supported through taxes levied indiscriminately upon all. Viewing these funds as public funds, the National Foundation has found it necessary to establish certain minimum standards. Realizing that professional staff and equipment are necessary, if poliomyelitis patients are to receive adequate treatment, the National Foundation has directed its chapters to pay for convalescent and long-term care only in those institutions that are approved by, or meet the standards of, the crippled-children's services of the states in which they are located. We recognize that this is a minimal standard, but it has served to eliminate certain institutions that are neither equipped nor staffed to meet the needs of the poliomyelitic patient. Since it is true that approved hospitals for crippled children are not everywhere equally available, the National Foundation, in conjunction with the crippled-children's services of the states, has attempted to select hospitals and to develop them as primary treatment centers. Poliomyelitis, being an epidemic disease, prevents one forecasting the potential case load in any single area in any given year; so that we have attempted, again in cooperation with the crippled-children's services, to develop in addition to the primary centers, secondary treatment centers that serve to receive the overflow, the idea always being that, as soon as facilities exist in the primary centers, the secondary centers will be closed. The adopting of these standards for hospitals has, in some measure, protected the chapters against paying for substandard service.

While we are not at all satisfied with the standards which have been established, we believe that we have done much to eliminate unqualified practitioners by adopting a policy where chapters may pay for medical service only to those physicians who are licensed to practice medicine and surgery.

The determination of those patients who shall receive financial assistance from the National Foundation chapters has been a difficult problem, a problem that has not yet been solved to the complete satisfaction of hospitals, doctors, and chapters. Recognizing that poliomyelitis, particularly when involvement is more than minimal, is a serious disease for which it may cost great sums of money to provide care, the National Foundation has felt that it should not give assistance only to those who are "indigent", but rather that it should use the Foundation funds as trust funds, making them available to persons who are "medically indigent". The family, by having these resources made available, is then protected from becoming truly indigent.

Although some of our chapters have viewed Foundation funds as a form of insurance—that is, a guarantee to all persons against any and all costs of poliomyelitis—this is quite contrary to the policy of the National Foundation and every attempt is being made, through education and publicity, to advise our chapter volunteers, as well as the public, that every patient with poliomyelitis has the responsibility to assume that portion of the cost of care which can be met from the patient's own resources.

The payment for services rendered poliomyelitic patients has constituted no great problem, with the exception of medical fees. Since various agencies, both public and private, are purchasing hospital care on a reimbursable cost rate, hospitals in this country

that are accepting poliomyelitic patients have cooperated, generally, in establishing a rate for poliomyelitic patients where the cost of care is being underwritten in whole, or in part, by a chapter. Fees for nursing and physical therapy again are well established, although they vary from state to state, and even within the state. The problem of reimbursing the physician for his professional service presents today one of the greatest problems to the National Foundation and its chapters. To place a dollar-and-cents value on a physician's professional ability is exceedingly difficult.

I wish that the National Foundation could arrive at an understanding with the physicians in this country as to how best to reimburse them for poliomyelitis care. The patient who can meet the entire cost of care from his own resources presents no problem. The indigent patient, for the most part, receives his medical attention in hospitals where medical service is provided through the staff of that institution. However, since poliomyelitis is no respector of an individual's economic status, there are many patients in the middle-income group who are not eligible for charity and who cannot meet the total cost of care from their own resources. Since the National Foundation is a public agency, recipients of National Foundation financial aid cannot be considered by the physician in the same category as the private patient when funds of the National Foundation are limited; and, in the light of the recurring epidemics of this disease, our chapters cannot be expected to pay for medical care at the same rate as physicians are charging their private patients. Equally true, as a voluntary agency limited in its ability to establish standards for medical care, it is not reasonable that our chapters should be expected to rate the quality of professional care and pay according to this rate. If chapters are to pay, it must be recognized that they shall have to pay the same fee to the qualified as well as to the unqualified physician. The simplest way to resolve this problem would be for the National Foundation to adopt as its policy that we would not pay for any medical service, but that the physician would have the right to collect his professional fee from his patient when caring for poliomyelitis as he would for any other illness. If the patient and the physician were advised of this, I am satisfied that, except in epidemic areas where patients are transported long distances to primary centers, the physician, in most instances, would stand an even greater chance of collecting his fee, since the National Foundation funds could be used for paying for hospital care, other professional services, such as physical therapy and nursing, and for the purchase of appliances.

The only other alternative to this is to establish in each state a predetermined fee schedule which must be adopted by the physicians and would be used by our chapters in those instances where the patient and the physician look to the chapter for the payment of the cost of medical care.

For many years poliomyelitis has constituted the greatest single cause of crippling in children. During the past four years, the incidence of poliomyelitis has more than doubled, and we have no reason to believe that the yearly average incidence will materially fall until such time as some means for controlling or preventing the disease has been established. The problem today is to see to it that patients with poliomyelitis everywhere have the same opportunity for optimal recovery. As stated before, the cost of providing this care in 1949 was in excess of $31,000,000. How much need be spent in 1950 cannot be estimated, and how much will be available for the care of poliomyelitic patients will depend upon the generosity of the American public and the ability of the National Foundation and its chapters to see to it that its funds are judiciously expended. Many of these patients require long periods of active treatment, which in the past has been carried out in hospitals. With hospital costs what they are today, every effort must be made to discharge patients early, referring them to physicians' offices and out-patient treatment centers. This is not a serious problem in our metropolitan centers, but poliomyelitis, being no respecter of the economic strata of society, is as likely to strike in the rural as in the urban centers. The poliomyelitic patient in the rural area is denied the greatest oppor-

tunity for recovery when discharged from the hospital without adequate medical supervision and follow-up. Every year thousands of dollars are being wasted by giving excellent care to the patient during the acute phase in the modern treatment center, when that patient is denied adequate follow-up. These are the patients who return years later with deformities that require repeated surgical procedures.

This medical-care program of The National Foundation for Infantile Paralysis should be the medical profession's responsibility. We look to you for counsel and guidance, just as our chapters should look to you as those best qualified to assist them in aiding the poliomyelitic patient. I know that we have made mistakes. We must recognize that not all of the $31,000,000 spent last year was spent wisely, but I am sure that you, as physicians, will help us in seeing that we make fewer mistakes, and that you will realize, at the same time, that we wish to work with you and through you.

QUESTION PERIOD

DR. H. R. MCCARROLL, ST. LOUIS, MISSOURI: Dr. Van Riper, among the officers of this organization you have many devoted friends. We have enjoyed your presentation and it gives us much hope.

I shall ask you a question, but it is very difficult to ask a question without first laying some preliminary ground work. In years past, our St. Louis chapter has followed a policy of giving expensive private medical care to all patients with poliomyelitis. As a result of this policy, the chapter ran out of money this year and came to the orthopaedic surgeons for advice. A committee appointed by the President of our local orthopaedic society studied this problem with the National Foundation Chapter and made two recommendations:

First, that every patient be transferred as soon as possible from the so-called primary treatment center to one of the clinics already established in the community where care could be given at no cost, or at minimal cost, to the Foundation Chapter.

Second, that prolonged physical therapy be discontinued unless recommended by a qualified orthopaedic surgeon.

I disapprove of any organization continuing to use public funds for expensive private medical care, when the patient can be cared for just as well on a more economical basis. My question, therefore, is simply this: Will The National Foundation for Infantile Paralysis be willing to sponsor such a recommendation and encourage a policy of this type among our local chapters, in order that some of this money can be saved for the continuation of valuable research which still offers our one hope for eventual solution of this problem?

DR. JOHN C. MCCAULEY, NEW YORK, N. Y.: I think Dr. Van Riper's paper has been very instructive. Certainly, the medical-care program of the National Foundation is comprehensive. I think its practical aspects are commendable, and the use of its financial reservoirs for aiding these poliomyelitis victims has been very generous.

I have several questions to ask Dr. Van Riper today: What does he believe to be the greatest deficiency in that program? What does he believe to be the greatest threat to his program? What does he think the practising physician should know about that threat or deficiency? What, in his opinion, can the practising physician do about it?

DR. J. ALBERT KEY, ST. LOUIS, MISSOURI: I did not know that the National Foundation had spent as much as $31,000,000. To me that is a lot of money.

It so happens that the orthopaedic surgeons have been to a large degree ignored in St. Louis, both in the policy making and in the activities of the local chapter. There are ample facilities in St. Louis for treating all of the patients who have infantile paralysis in a manner which we believe will give the best end results, but not necessarily in a manner which the advisors of the local chapter believe will give the best end results.

The Foundation should discontinue the practice of insisting to parents of patients, who are well-to-do and quite able to take care of their medical bills and hospital bills for as long as they need care, that these patients be turned over to the National Foundation for care, claiming that they alone can give the treatment that will prevent crippling. No one has yet produced evidence that the orthopaedic surgeons of this country are incapable of treating poliomyelitis adequately.

As the committee of St. Louis orthopaedic surgeons told the National Foundation, our local orthopaedic surgeons are more than glad to render free care to patients who are deserving of free care. We do not want to charge people who cannot pay. At the same time, we do not think that the National Foundation should pay for the care of people who can afford to pay, nor do we think that they should continue to support prolonged and unnecessary hospitalization and physical therapy. Much of their substance has been wasted in this manner.

DR. HAROLD E. CROWE, LOS ANGELES, CALIFORNIA: I am very glad to ask this question because it was suggested to me recently by a course on the treatment of poliomyelitis, which was presented at the Los Angeles Orthopaedic Hospital to a group of doctors from outlying areas. In the sessions of four days, the program was largely devoted to teaching that each patient with poliomyelitis must have an orthopaedic surgeon, a physical therapist, a social worker, and a psychiatrist.

Being the only dissenter, I asked the question when and how should one stop treatment for the best good of the child, without hurting the child, and without allowing the youngster to feel abandoned by his physician. This problem seems to me one of the most important decisions to make in the treatment of anterior poliomyelitis.

I feel that the National Foundation is such an important organization that we cannot afford to let it dissipate its funds in unnecessarily refined treatment as happened in the Los Angeles area in the past year. We have a great need for funds to care for the young adult patient who, at the present time, has difficulty in finding satisfactory facilities for treatment. We have adequate facilities in Los Angeles for the care of children suffering from this disease, but have very few satisfactory arrangements for the young adult. This discrepancy in our set-up has not been handled adequately by the National Foundation to this time.

Consequently, with a desire to aid the Foundation and conserve its funds, I wish to call attention to the problem of endless expenditure on unnecessary treatment, pointing out that it is very easy to get into a rut, so that treatment goes on indefinitely with stretching, muscle training, and physical therapy. The Foundation, for its own financial good, should find an answer to the question of how best to stop such treatment and to direct the child into the new life which he must some day face and to which he must adjust himself with his new handicap, irrespective of how severe that handicap may be. The lives of many handicapped persons are wasted in endless, useless treatment, when these people might much better be restored to some active and productive position in the community in spite of a persistent and incurable physical disability. How can we get out of this rut?

DR. HART E. VAN RIPER, (close of question period): In answer to Dr. McCarroll's question: After the overwhelming epidemic of 1949, The National Foundation for Infantile Paralysis and its approximately 2,800 chapters must conserve to the fullest the funds that are available for patient care, to the end that no patient be denied the needed treatment. One of our greatest problems today is over-hospitalization and continued treatment beyond the point where the patient actually continues to benefit. It must be recognized that many patients in urban centers can be discharged from the hospital and followed in out-patient clinics or physicians' offices without detriment to the patient. However, such out-patient service is not equally available to children from rural areas. Through proper planning of field services with crippled children's agencies, it may be possible to institute earlier hospital discharge even for this rural group.

I am certainly of the opinion that physical therapy should be given only under the direction of a competent physician experienced in poliomyelitis, who has an understanding of the rehabilitation of the physically handicapped. I am convinced that many patients are receiving physical therapy far beyond the point where they are benefiting.

Answering Dr. McCauley's question: It is a policy of the National Foundation that every one of its chapters must have a Medical Advisory Committee if that chapter is to be properly organized for service to poliomyelitic patients. If it were possible to have every single physician in the United States who has contact with poliomyelitic patients understand the program of the National Foundation and its policy of helping financially only those patients who need help, then I am sure that the medical profession as a whole would make an even greater contribution to the National Foundation's program. We believe that the attending physician is the one best qualified to determine his patient's needs. If every physician would look upon National Foundation funds as he would upon the family's funds and would attempt to conserve these funds, much could be accomplished in the reduction of the costs of patient care while, at the same time, the patient would be assured of the greatest possible recovery. When one views the increasing incidence of poliomyelitis over the past few years, one must recognize the need to utilize these funds in as conservative a way as possible. Since the practising physician is the one who orders treatment, that physician is the one who can do most to assist the National Foundation in its medical-care program.

In reply to Dr. Key's question: I am happy to say that the St. Louis situation is not so acute as it was a short time ago. The cooperation of the orthopaedic surgeons in St. Louis is most commendable, and I believe that within the coming months we will find considerable improvement in chapter policy and function.

Replying to Dr. Crowe's question: I believe that Dr. Crowe misunderstands the program that is being presented at the Los Angeles Orthopaedic Hospital. The millenium will have arrived when it is possible for every child to have, in addition to good medical supervision, adequate physical therapy, occupational therapy, and medical social work, as well as psychological understanding of the problems of the physically handicapped. The program at the Los Angeles Orthopaedic Hospital is an attempt to develop teamwork within an institution, and to develop such teamwork, it is necessary that every member of the team understand the contribution that other members of the team have to make to the total patient-care program.

VITAMIN-RESISTANT RICKETS*

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In crippled-children's clinics, it has long been known that some cases of infantile rickets do not respond to the usual treatment with vitamin D but will respond to larger doses. Therefore, many cases of moderately resistant rickets have been treated adequately with from 10,000 to 20,000 units of vitamin D daily. It was not until 1937, however, that Albright [1] described a much more severe and vitamin-resistant type of rickets, which he called vitamin-resistant rickets. He had then seen six cases and he presented a complete metabolic study of one. He pointed out that the disease differs from infantile rickets only in its severity, its failure to respond to the usual treatment, and its normal response to massive doses of vitamin D (100,000 to 1,500,000 units daily). In the untreated state, the disease is characterized by short stature and the usual physical and roentgenographic findings of florid rickets. Blood studies show decreased inorganic phosphorous, normal or low serum calcium, increased serum alkaline phosphatase; other findings are decreased urinary calcium, and increased faecal content of calcium and phosphorous. With these findings, there is no evidence of kidney, liver, or gastro-intestinal disease.

Albright's metabolic studies, which included the use of intravenous injections of vitamin D, indicated that vitamin D was absorbed, but that there was no response in the patient until a threshold level had been reached; above this level the response was normal. Signs of toxicity appeared, however, if the vitamin-D intake was too great. An adequate response by the patient was indicated by increased urinary calcium, and decreased faecal calcium, and in the blood by increased inorganic phosphorous, normal or elevated calcium, and a gradual decline in alkaline phosphatase. Accompanying that chemical response was roentgenographic evidence of healing and growth of the patient at the rate of one centimeter a month. Albright also pointed out that once healing had been established, the maintenance dose of vitamin D might be markedly decreased.

Since that report, individual case reports [4,5,6,7,8,10,12,13,14], and discussions of the physiology involved have served to establish vitamin-resistant rickets as a distinct clinical entity. These reports have apparently established the fact that vitamin D is absorbed at the usual dosage and that it may be present in the blood stream at concentrations many times in excess of the normal without evidence of chemical or roentgenographic response [4,6]. Each report has emphasized that an adequate dosage must be established for each patient by trial and error. When an adequate response has been finally secured, the maintenance dosage for each patient must be determined in a similar manner. Usually, this will be found to be approximately one-half to one-third of the initial requirement. Thereafter, each patient must be followed carefully by means of urinalyses and blood determinations of calcium and inorganic phosphorous, in an effort to prevent overdosage with subsequent toxicity, metastatic calcification, and even irreparable kidney damage.

Some of the reported cases indicate a familial incidence. Christensen, McCune, Gill, and Kajdi have each reported a family group with this type of involvement. Case reports of familial "late" rickets have been made by Jampolis and Londe and Lightwood and Wood. The latest report by Freeman and Dunsky indicates that this is a very rare disease, and that no more than thirty cases have been reported. However, the present study indicates that the condition is much more common than has been generally realized, that

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TABLE I

FAMILY INVOLVEMENT

F. R.	R. L.
Grandfather	S. L.
Mother	B. L.
3 Aunts	Mother
3 Cousins	
J. D.	B. F.
V. D.	L. F.
Great grandmother	Father
Grandmother	T. H.
Great aunt	B. H.
Mother	Mother
B. B.	A. S.
Grandmother	Father
Father	
Uncle	C. B.
	Mother
L. W.	
J. W.	G. C.
Mother	Father

The sixteen children with proved familial characteristics are listed by initials, in their respective family groups. The immediate parents listed, with one exception, have been recently examined. The remaining members of families were accepted on the history obtained from the parents.

there are varying degrees of severity, and that there is a very marked familial tendency. It was because of this obvious familial characteristic that the authors became interested in further detailed study of this disease.

This investigation deals with a series of twenty-five such patients who have been treated at the St. Louis Unit of the Shriners' Hospital for Crippled Children, and represents the number of such patients seen since 1933. All had a history of rickets beginning in infancy. Each had been treated with large doses of vitamin D, but, in spite of this, florid rickets was still demonstrable by roentgenograms. This original treatment had usually consisted of twenty to seventy-five drops of a cod-liver-oil concentrate daily, or cod-liver oil fortified with percomorph oil, one teaspoonful three or four times daily. None of the twenty-five children had any evidence of an abnormal dietary history or of gastrointestinal disease, and all had several normal urinalyses and blood counts. In each instance, however, the presence and persistence of deformities had necessitated one or more osteotomies in the lower extremities.

Eight of these children were admitted to the Hospital for re-examination and study, while the others were called for study and treatment as out-patients. The routine study of each included the following: a complete blood count, urinalysis (including the Sulkowitch test), urine concentration test (unless random specimens revealed a specific gravity of 1.025 or better), serum calcium, inorganic phosphorous, and alkaline phosphatase, plasma chlorides, plasma carbon-dioxide content, thymol turbidity, fasting blood sugar, total and fractional plasma proteins, and non-protein nitrogen determinations. Complete studies have now been obtained on eighteen of the twenty-five patients. Of the remaining seven, two have had all tests except the determination of plasma carbon-dioxide content. Three patients were included in the study on the basis of their history, physical examination, and the roentgenographic findings. Urinalyses and blood determinations for calcium, inorganic phosphorous, and alkaline phosphatase revealed levels characteristic of rickets. The remaining two patients were included in the study solely on the basis of history, physical examination, roentgenographic findings, and urinalyses. Of the twenty-five children, seventeen are still being treated with massive doses of vitamin D, ranging from 100,000 to 600,000 units daily. The preparation used, "ertron", is an activated vaporized

TABLE II

CLINICAL AND CHEMICAL FINDINGS

ADULTS

Patient	Calcium (*Milligrams per 100 cubic centimeters of serum*)	Phosphorous (*Milligrams per 100 cubic centimeters of serum*)	Alkaline Phosphatase (*Bodansky units*)	Height (*Feet*)	(*Inches*)
G. R.	9.7	2.1	3	4	11
	9.6	0.7	3		
A. D.	9.9	1.9	5	4	6
	9.1	0.8	6		
A. S.	9.2	0.7	4.7	4	8
	10.1	2.5	3		
L. C.	10.1	0.5	6	5	¼
A. B.	11.4	1.8	4	5	5¾
F. B.	8.8	1.7	3	4	6¼
L. W.	10.4	4.2	2	4	3½
L. L.	9.7	3.7	4	4	7
M. H.	11.6	1.9	3	4	3¾
T. H.	10.2	1.3	3	4	8¼

Pertinent clinical and chemical findings in the ten adult patients examined. All but two showed a low serum phosphorous, and all but two are below five feet in height. The serum calcium and alkaline phosphatase levels were within normal limits.

ergosterol, dispensed in capsules which contain five milligrams of activation products having an antirachitic activity of 50,000 U.S.P. units.

It was found that sixteen of the twenty-five patients fell into ten family groups, in which one parent in each group presented similar deformities and characteristic histories. Nine of the ten parents have been adequately studied, and five of them are under treatment with 300,000 units daily at this time. At this Hospital three of the parents, as children, had had osteotomies for the correction of deformities due to rickets. (They are not included in the present series of twenty-five cases.) Two of the parents had had similar treatment at other hospitals; four had had no such treatment, although three of these now have severe deformities. Histories concerning other members of the family were obtained from the parents who were able to tell of still others who had similar short stature and deformities. This material is recorded in Table I.

Several striking features were revealed in the examination of these patients. They presented the usual signs of active rickets, such as the enlargement of all the palpable epiphyses, Harrison's groove, rachitic rosary, and frontal bossing. In addition, there were certain features in all which had presumably resulted from the long-standing florid rickets. The most marked of these was short stature which results primarily from the lack of growth, varying with the severity of the disease, in the lower extremities. This feature is especially striking in adults. Of the ten adults examined, the height of one was found to be five feet, five and three-quarters inches, and of another to be five feet and one-quarter of an inch, while the others were below five feet in height. This is recorded in Table II. Another noticeable feature was the presence of severe deformities in the lower extremities,—bow legs, knock-knees, or the "tackle deformity" which is a bow on one side and knock-knee on the other. In several of the more severe cases, there was also a marked anterior bow of the femora and tibiae. It is characteristic of these deformities that they tend to recur after early correction by osteotomies. The group of twenty-five children has had 122 osteotomies, twenty-five of which were for recurrences.

Although the exact nature of this disease entity has only recently been recognized in this Clinic, it has long been known that most deformities caused by rickets recur after early correction. On that basis, correction has often been delayed until the age of epiphyseal closure in an attempt to decrease the number of osteotomies required for recurrences. On the other hand, the follow-up in this Clinic is limited to fifteen years of age and a recent examination of adults proves that late recurrences may still occur. Associated with these severe deformities, there is ligamentous instability about the joints, seen most commonly at the knee. These patients with ligamentous instability frequently complain of pain on moderate exertion and have occasional episodes of severe pain. Also associated with the deformities, is a characteristic waddling gait, due to a fairly marked coxa vara.

Characteristic deformities are also common in the skull. There is an increased anteroposterior diameter, with a decreased transverse diameter. In addition to the usual frontal bossing, there is often a marked external occipital protuberance. A typical saddle nose may be present. Dental examinations have been conducted in eight of these patients, but the dentist has been unable to make a definite statement of the effect of rickets upon teeth.

The roentgenographic evidence of such severe rickets may be classified under three headings: changes in the general architecture of bone, characteristic alterations of the epiphyseal structures, and deformities. In the cancellous bone of metaphyseal areas, the trabeculae are coarser, broader, and more widely spaced than usual. The epiphyseal lines are much wider than usual. On the metaphyseal side of the epiphyseal line, the zone of calcification may be even and abrupt; but the usual finding is one of irregular calcification, presenting a "wooly" or fuzzy appearance. An interesting and constant roentgenographic finding has been some improvement in the appearance of the epiphyseal lines following postoperative plaster immobilization. This improvement was noted in every patient who had had adequate postoperative roentgenograms, and the effect was general, not local. Available roentgenograms, unfortunately, include only the lower extremities, but all the visible epiphyses improved to the same degree, regardless of which bone was osteotomized. It should be noted that, of all the epiphyses, improvement is easiest to see in the upper tibial epiphyses, whether it be the result of plaster immobilization or of vitamin therapy.

FIG. 1-A

A.S. This patient is characteristic of the adults who appear to have active disease. The relative shortening of the lower extremities and the recurrence of deformity is typical. As a child, he had osteotomies for the correction of deformities due to rickets.

The results of laboratory studies in every instance have been those characteristic of rickets, with no other abnormalities demonstrable. In twenty-three patients on whom adequate blood-chemistry studies were done, the maximum inorganic phosphorous level was found to be 3.3 milligrams per 100 cubic centimeters of serum, while in twenty patients the minimum was found to be below 3 milligrams per 100 cubic centimeters of serum. The serum calcium in all cases was normal or slightly low. The serum alkaline phosphatase was elevated to 20 or more Bodansky units in twelve patients, was between 15 and 20 in four patients, and was 12 units or less in seven patients. The youngest of the latter seven patients was thirteen years of age. In other words, each of these patients had arrived at the age of diminishing epiphyseal activity. In every case, the urinalysis was within normal limits; the Sulkowitch test was negative to a bare trace, as opposed to a one to two plus reaction for normal. The urine concentration tests were within normal limits, and each patient demonstrated the ability to excrete an acid urine. There was no evidence of acidosis, disturbed carbohydrate metabolism, or liver disease.

FIG. 1-B

FIG. 1-C

FIG. 1-D

Fig. 1-B: A.S. Characteristically, there are degenerative changes about the hips and lumbosacral spine. There is definite coxa vara and an increase in bony mass at the muscle attachments (greater and lesser trochanter). Also visible are Looser's lines in the femora.

Fig. 1-C: A.S. In the adult spine, there are biconcave vertebrae and biconvex disc outlines, with extensive degenerative arthritic changes. The concave vertebral borders are also seen in the anteroposterior view. Note in this film, the mild compression of the twelfth thoracic vertebra with no history of injury.

Fig. 1-D: A.S. In the upper extremities of adults the long bones are short and thick and there is enlargement at the major muscle attachments.

Special mention should be made of the findings in adults. The degree of dwarfism, the normal alkaline phosphatase, and the variable inorganic phosphorous levels can be noted in Table II. One of this adult group is also included in the group of twenty-five. He is now twenty-six years of age but was originally treated here as a child. His younger sister is now under treatment, and his mother is one of the adult patients examined. All adults complain of marked fatigability and inability to do hard work. They complain particularly of low-back pain and pain in the knees and hips. Six of the group are known to have had progression of their deformities since the age of epiphyseal closure, accompanied by instability about the knees (Fig. 1-A). By roentgenographic evidence, six of the adults have the same typical, coarse architecture of bone, which is noted in children. Two showed definite Looser's lines (Fig. 1-B); one of these showed symmetrical lines on the medial border of the upper third of each femur, while the other showed a comparable change in the upper third of the medial aspect of one tibia. Their vertebrae are characteristically biconcave in both views, with biconvex disc outlines (Fig. 1-C). Those who present the greatest deformities show also marked degenerative changes about the knees, hips, and lumbosacral spine. At the hip joint, increased depth of the acetabulum and coxa vara have been noted. There is an increased bony mass at the site of the attachments of the major muscles, such as the greater and lesser trochanters and the deltoid insertion on the humerus (Figs. 1-B and 1-D).

In the attempted study and management of this series of twenty-five patients, two plans of treatment were established:

The first plan dealt with hospital patients who were treated vigorously in an effort to determine the maximum safe response and to elevate the serum phosphorous levels to normal. When the maximum response, or toxicity, had been elicited, treatment was discontinued to study the pattern of regression. The patients were followed by monthly determinations of their height and weight, and weekly urinalyses and blood-chemistry studies. Roentgenograms were first taken at weekly intervals, but are now taken monthly.

The second plan was developed for out-patients who were treated with smaller doses of vitamins and checked at monthly intervals by means of urinalyses, blood-chemistry determinations, and roentgenograms. This plan of treatment can be justified by the established fact that healing, as determined by roentgenogram, and increased urinary calcium excretion, occurs in the absence of appreciable elevation of inorganic phosphorous. Treatment was empirically started with 300,000 units daily, with revisions upward or downward, depending upon the effect desired. The dosage in out-patients is increased or decreased to maintain a Sulkowitch test of one to two plus, at which level calcium casts do not usually appear. Calcium is not usually appreciably elevated. With hospitalized patients, however, the dosage was increased until the inorganic phosphorous reached 5 milligrams per 100 cubic centimeters or until toxic symptoms appeared. In this group the Sulkowitch test was allowed to reach four plus for short periods if necessary.

The response to treatment by different patients is extremely variable and serves to emphasize the statement that each patient is an individual problem. All of the patients treated have shown typical chemical and roentgenographic changes. On adequate dosage with vitamin D, the serum inorganic phosphorous and urinary calcium rise, and the alkaline phosphatase gradually declines. Changes in urinary calcium and serum inorganic phosphorous may be seen within a week, while roentgenographic changes do not appear for three or four weeks. In seven patients, a definite attempt was made to elevate the inorganic phosphorous to 5 milligrams per 100 cubic centimeters of serum. That was possible in each case; however, maintenance of an inorganic phosphorous level above 4 milligrams per 100 cubic centimeters of serum led to gradually increasing serum calcium levels, a four plus Sulkowitch test, the presence of many calcium casts in the urine, and the

FIG. 2

This shows a typical family group with the involved children, from left to right, being six (Figs. 5-A to 5-F), seven (Figs. 4-A to 4-E), and eight (Figs. 3-A to 3-H), years of age. The fourth child who is nine years of age and four other children have no demonstrable disease. The mother is four feet and seven inches tall and, as a child, had eight osteotomies for the correction of bow legs. Each of the three involved children has had osteotomies for correction of lower-extremity deformities.

clinical signs of toxicity. The clinical signs of toxicity are: anorrhexia, nausea, vomiting, weight loss, occasionally hematuria, and rarely oliguria or anuria associated with nephrocalcinosis. Toxicity, while on massive vitamin-D therapy, has not been seen in this group in the absence of definite hypercalcaemia; it has not been seen in a patient with a calcium level below 12 milligrams per 100 cubic centimeters of serum, and a urine Sulkowitch test below three to four plus. The urine during toxic states contains many calcium casts. Only one instance of gross hematuria and oliguria has been encountered, and that occurred with calcium levels below 12 milligrams per 100 cubic centimeters of serum. Vitamin D was stopped, fluids were forced; the hematuria and oliguria disappeared in two days.

Seven patients in the series were treated with massive doses of vitamins during plaster immobilization following osteotomies. That treatment occurred early in this study, but without serial chemical studies. All thus treated showed very marked improvement at the epiphyseal lines, but all developed severe anorrhexia, nausea, vomiting, and weight loss. It has been demonstrated by Albright [1], and others, that following plaster immobilization in a young active child there may be an increase in the serum calcium and phosphorous. Such an increase, in addition to that resulting from massive vitamin therapy, may lead to serious toxicity. Three of these seven patients have had subsequent prolonged treatment with the same vitamin dosage, but without immobilization. These have shown no elevation of serum calcium or evidence of toxicity. When massive vitamin therapy was discontinued at the peak of chemical response, there followed, within one week, a decrease in serum calcium, serum inorganic phosphorous, and urinary calcium excretion. The drop to the

FIG. 3-A

An increased anteroposterior skull diameter and occipital bossing is common in children with severe long-standing rickets.

FIG. 3-B

pretreatment level was usually complete in two weeks or, occasionally, three weeks. Unless vitamin D was omitted for more than one month, no change was noted on the roentgenograms.

Roentgenographic evidence of healing is seen at the epiphyseal line. It is actually demonstrable at all epiphyses, but for purposes of demonstration and for uniformity the knee joint alone will here be discussed. It is always true that, at the knee joint, improvement is best seen at the upper tibial epiphysis. Along with healing, there is also seen evidence of a progressive calcification beginning at the metaphyseal side of the epiphyseal line and progressing toward the epiphysis itself. In other words, the first evidence of healing

FIG. 3-C

At four years of age there was bowing, and wide, irregular epiphyseal lines and coarse architecture of metaphyseal bone.

is the calcification of a wide area of uncalcified osteoid tissue and cartilage which has accumulated at the epiphyseal line. That area of calcification increases in density and progresses toward the epiphysis until a normal, narrow, regular epiphyseal line is seen, and until mineralization in the metaphysis is homogeneous. No patient has been treated long enough to demonstrate alteration in the appearance of the general architecture of bone.

For a more accurate demonstration of the typical findings, plan of treatment, and the response thereto, three children of a single family group (Fig. 2) are reported in detail:

R. L., X-92, was first seen at four years of age with bilateral bow legs, first noticed at eleven months of age when he began to walk. He had been given cod-liver oil and orange juice, beginning at the age of two months. His mother was four feet and seven inches tall and had had eight osteotomies for similar deformities as a child. Two younger sisters had similar deformities. His only previous illness was uncomplicated measles. A physical examination revealed bilateral bow legs, with the bow involving both femora and tibiae. There was also some anterior bowing in each tibia. There was a Harrison's groove, dolichocephaly (Figs. 3-A and 3-B), and an enlargement of all palpable epiphyses. Roentgenograms revealed the typical deformities and epiphyseal changes characteristic of rickets (Fig. 3-C). Treatment as an out-patient consisted of 3,500 units of vitamin D daily. There was no improvement in the appearance of the bones by roentgenogram or in the deformities (Fig. 3-D). He was admitted to the Hospital for bilateral supracondylar osteotomy of the femur and bilateral osteotomy of the tibia. This necessitated immobilization for a period of several months, in which there was a distinct improvement in the appearance of the epiphyseal lines (Fig. 3-E). He was then discharged, was kept on forty-five drops of cod-liver-oil concentrate daily, and was followed again as an out-patient. In spite of this therapy, there was regression to florid rickets within a period of a few months (Fig. 3-F), and he was re-admitted to the hospital for study. His laboratory findings were as follows: no abnormality in blood count or in urinalysis, Sulkowitch test was 0 to +, urine concentrated to 1.030, calcium 11.2 milligrams per 100 cubic centimeters of serum, inorganic phosphorous 2.4 milligrams per 100 cubic centimeters of serum, alkaline phosphatase 28 Bodansky units, plasma chlorides 387 milligrams per 100 cubic centimeters of serum, and carbon-dioxide content 62.5 volumes. No abnormalities were found on determination of the fasting blood sugar, non-protein nitrogen, total and fractional plasma proteins, and thymol turbidity. Massive vitamin-D therapy was instituted. The chemical response to 450,000 to 600,000 units of vitamin D daily, and the reaction to its withdrawal is recorded in Figure 3-H. There was a prompt rise of calcium to 12.8 milligrams per 100 cubic centimeters of serum, and of inorganic phosphorous to 4.5 milligrams per 100 cubic centimeters of serum, accompanied by increased urinary excretion of calcium to a four plus by Sulkowitch test. At that point, he had mild clinical toxicity and showed calcium casts in the urine. The prompt reversal of these effects was noted on withdrawal of vitamin D. After four months of massive vitamin therapy, his maintenance dose was established at 150,000 units daily and there was roentgenographic evidence of healing (Fig. 3-G).

FIG. 3-D

After fifteen months of treatment, with at least 3,500 units of vitamin D daily, the epiphyseal lines were still wide and irregular and the deformities had increased.

B. L., Z-71, sister of R. L., was first seen at the age of five years with a complaint of knock-knees. The deformity had progressed since sixteen

months of age, when she began walking. Five drops of cod-liver-oil concentrate had been given daily from early infancy. Her past history included only measles and whooping cough. Physical examination revealed

FIG. 3-E

Unfortunately this x-ray shows bone detail very poorly but, if studied closely, considerable improvement in the appearance of the epiphyseal lines can be noted. This change resulted from postoperative plaster immobilization.

FIG. 3-F

FIG. 3-G

Fig. 3-F: Six months after plaster immobilization, there was regression to active rickets, in spite of an intake of 9000 units of vitamin D daily.

Fig. 3-G: Prompt healing was noted at the epiphyseal lines after four months of massive vitamin therapy.

an enlargement of all palpable epiphyses, dolichocephaly, Harrison's groove, and severe genu valgum. Roentgenograms revealed the typical deformities and wide irregular epiphyseal lines of rickets (Fig. 4-A). She was treated with approximately 9,000 units of vitamin D daily for six months with no improvement in the physical findings or in the roentgenograms. She was then hospitalized and had bilateral supracondylar osteotomy of the femur, followed by bilateral osteotomy of the upper third of the tibia. For two and one-half months following the supracondylar osteotomies she was given 300,000 units of vitamin D daily. This combination of immobilization and massive vitamin therapy produced signs of marked toxicity. Roentgenograms obtained after the second osteotomies, however, revealed the epiphyseal lines to be normal in appearance (Fig. 4-B). She was then treated as an out-patient on 9,000 units of vitamin D daily for three months, at which time the wide epiphyseal lines were again characteristic of florid rickets (Fig. 4-C).

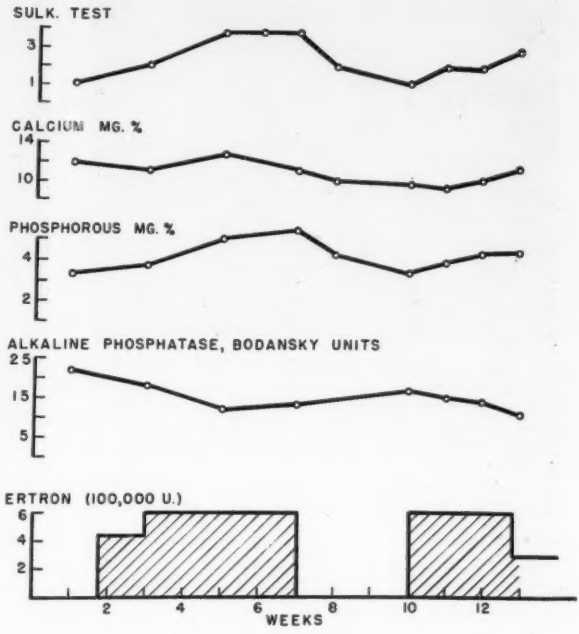

FIG. 3-H
Vitamin-D therapy and chemical response in patient R.L.

She was again hospitalized and the following laboratory data were obtained: no abnormalities in blood counts or urinalyses, Sulkowitch test 0 to +, urine concentrated to 1.033, calcium 10.8 milligrams per 100 cubic centimeters of serum, inorganic phosphorous 2.0 milligrams per 100 cubic centimeters of serum, alkaline phosphatase 18 Bodansky units, plasma chlorides 390 milligrams per 100 cubic centimeters and carbon-dioxide content 48.5 volumes. The fasting blood sugar, non-protein nitrogen, thymol turbidity, and total and fractional plasma proteins remained within normal limits. Her subsequent response to massive doses of vitamin D is summarized graphically in Figure 4-E. With treatment, there was increased calcium excretion in the urine, elevation of the serum calcium and phosphorous, and depression of the serum alkaline phos-

FIG. 4-A

FIG. 4-B

Fig. 4-A: Prior to treatment, the characteristic changes of rickets were noted: wide, irregular epiphyseal lines, coarse metaphyseal bone, obvious knock-knee.

Fig. 4-B: Following combined postoperative plaster immobilization and massive vitamin-D therapy (300,000 units daily), there was marked improvement in the appearance of the epiphyseal lines. During the period, however, there was clinical evidence of marked toxicity.

FIG. 4-C

Fig. 4-C: Three months after massive vitamin-D therapy and immobilization were stopped, there was again evidence of very active rickets. During that three-month period, the patient received 9000 units of vitamin D daily.

FIG. 4-D

Fig. 4-D: There was marked improvement in the appearance of the epiphyseal lines after four months of massive vitamin-D therapy.

phatase. When treatment was discontinued, the blood chemistry showed regression within one week, and with restitution of adequate vitamin-D therapy, it showed improvement within one week. There was marked roentgenographic evidence of healing after an additional four months of massive vitamin therapy (Fig. 4-D). She is now on 200,000 units of vitamin D daily.

S. L., Z-6, sister of R. L. and B. L., was first seen at the age of three years, also because of bilateral knock-knees. She walked at eleven months of age, and six months later her parents noted the deformities, which had been progressive. She had received five drops of a cod-liver-oil concentrate daily, beginning at the age of six weeks. Her past history was non-contributory. Physical examination revealed moderate, bilateral knock-knee, dolichocephaly, enlargement of all palpable epiphyses, and short stature. Roentgenograms revealed florid rickets. She was treated as an out-patient with 9,000 units of vitamin D daily in the form of a cod-liver-oil concentrate. There was no improvement clinically or as shown by roentgenograms after two years (Fig. 5-A), and she was hospitalized for bilateral supracondylar osteotomy of the femur, and bilateral osteotomy of the tibia. During her plaster immobilization, she received 300,000 units of vitamin D daily for four months. During this period, she had definite toxicity as evidenced by anorrhexia, nausea, vomiting, and weight loss, but roentgenograms revealed the rickets to be healed (Fig. 5-B). With treatment again reduced to 9,000 units of vitamin D daily, as an out-patient for three months, florid rickets again became evident by roentgenogram (Fig. 5-C). At this point, the following laboratory data were obtained: normal blood counts and urinalyses, Sulkowitch test 0 to

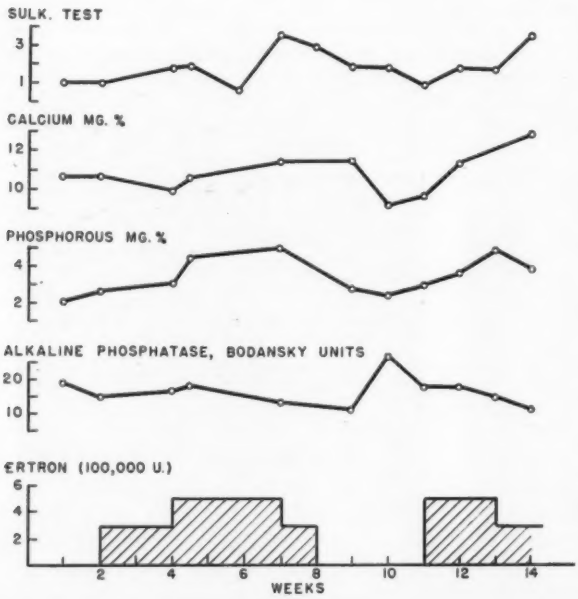

FIG. 4-E

Vitamin-D therapy and chemical response in patient B.L.

+, urine concentrated to 1.027, calcium 10.5 milligrams per 100 cubic centimeters of serum, inorganic phosphorous 2.1 milligrams per 100 cubic centimeters of serum, alkaline phosphatase 15 Bodansky units, plasma chlorides 380 milligrams per 100 cubic centimeters of serum, and carbon-dioxide content 65 volumes. The fasting blood sugar, non-protein nitrogen, thymol turbidity, and total and fractional plasma proteins were normal. She has since been treated with 300,000 to 600,000 units of vitamin D daily, both as an in-patient and out-patient. Her chemical responses are demonstrated graphically in Figure 5-F and her recent improvement shown by roentgenogram, is demonstrated in Figures 5-D and 5-E. On massive vitamin-D therapy her response has been satisfactory. It should be noted that a vitamin dosage which produced severe toxicity during plaster immobilization (300,000 units daily) was well tolerated for three months as an out-patient, with no blood chemistry changes, but with roentgenographic evidence of improvement. Also, during a second hospital admission, she showed clinical toxicity on 600,000 units daily when the serum calcium rose to 16.6 milligrams per 100 cubic centimeters. The urine at this time contained many calcium casts and showed a four plus by Sulkowitch test. She is now receiving 300,000 units daily.

DISCUSSION

In this study, no attempt has been made to establish vitamin-resistant rickets as a disease entity, since it has already been well described. It is severe rickets which responds only to massive doses of vitamin D. There is no evidence of gastro-intestinal, pancreatic, liver, or kidney disease. It is differentiated from renal acidosis and the Fanconi syndrome by decreased urinary calcium excretion, the absence of glucose, albumen, and amino acids in the urine, and the absence of disturbed plasma electrolytes.

The site of the metabolic error has been discussed in several reports. Albright [1] demonstrated that there was no response to usual quantities of intravenous vitamin; and, by determination of the blood levels of vitamin D, others have shown a lack of response at blood levels many times normal. Bakwin suggested that like carotene, vitamin D might have to be converted into some other substance before becoming active. Another group of workers has indicted the renal tubule as the site of disease,—they have shown increased phosphorous excretion in the untreated state and increased tubular re-absorption after treatment. On that basis, this disease is related to renal acidosis due to tubular disease and the Fanconi syndrome. However, definite parathyroid hyperplasia is known to exist in rickets, and Albright points out that the increased phosphorous excretion in rickets is probably due to parathyroid activity. As vitamin therapy increases the serum-calcium level, there should be decreased parathyroid activity and, therefore, decreased phosphorous excretion. Albright feels that there is a threshold effect in vitamin-resistant rickets. There

FIG. 5-A

FIG. 5-B

Fig. 5-A: Florid rickets and severe deformities present after twenty-one months of treatment with 9000 units of vitamin D daily.

Fig. 5-B: Following four months of combined postoperative plaster immobilization and massive vitamin-D therapy (300,000 units daily), there was marked improvement in the appearance of the epiphyseal lines. However, there was marked clinical toxicity during this period.

is no response to vitamin D until the threshold is reached. Above that level, the response is normal.

Whatever the basic defect, treatment is based on the threshold concept. Separate determinations of the threshold level and the level of toxicity must be made for each patient. The effect of long continued treatment with such massive doses is still unknown; Bakwin gave one patient 440,000 units daily for fifteen months with no deleterious effect. It is known that the presence of a few calcium casts in the urine is not harmful; it is also known that, when many calcium casts or hematuria appear, the effect is rapidly reversible by discontinuing treatment and forcing fluids. Albright emphasizes the danger of hypercalcaemia, and points out that, if serum levels above 12 milligrams per 100 cubic centimeters persist, nephrocalcinosis may result, and that, if the level is above 16 milligrams per 100 cubic centimeters of serum, generalized metastatic calcification may result.[3] Experience, derived from this study, confirms his conviction that all toxic effects are due to hypercalcaemia.

Since there is a relatively narrow margin between the threshold level and the level of toxicity, it appears that the logical long-term dosage schedule should be determined by urinary calcium excretion, serum alkaline phosphatase levels, and roentgenographic evidence of healing. Healing will occur when the Sulkowitch test is consistently one to two plus, even though the product of the serum calcium and phosphorous is close to thirty. If at that level there is a gradual decline of serum alkaline phosphatase and progressive healing is shown by roentgenogram, a satisfactory level has been reached. Monthly or bimonthly determinations of the serum calcium and phosphorous are important, however; for tolerance to a constant vitamin dosage for a period of two months has been observed with no effect on serum calcium, only to be followed by a gradually increasing calcium level to a state of toxicity.

FIG. 5-C

Three months after immobilization and massive vitamin therapy had been discontinued, there was regression to active rickets in spite of the use of 9000 units of vitamin D daily.

The strong familial character of this disease is striking, and it is partly because of this relationship that the disease has often been wrongly diagnosed in this Clinic. The disease appears as a familial dwarfism, with most of the shortening located in the extremities. Although epiphyseal changes suggesting rickets were observed, there was no response after supposedly more than adequate vitamin therapy. These conditions in the past, therefore, have been incorrectly classified as achondroplasia, dyschondroplasia, chondrodysplasia, or combinations of these diseases with rickets. The diagnosis of vitamin-resistant rickets in such patients can be definitely estab-

FIG. 5-D

Fig. 5-D: After three months, while ambulant as an out-patient, receiving 300,000 units of vitamin D daily, there was definite, progressive calcification of the wide epiphyseal lines. During this period, the patient was asymptomatic and gained weight. The same daily vitamin intake during immobilization, however, had produced marked toxicity.

FIG. 5-E

Fig. 5-E: After seven months of massive vitamin therapy (300,000 to 600,000 units daily), there was marked improvement in the appearance of the epiphyseal lines.

lished by urine and blood-chemistry studies. It represents the most common cause of dwarfism in this Clinic.

Two problems now become apparent: Can dwarfism be prevented, and can deformities

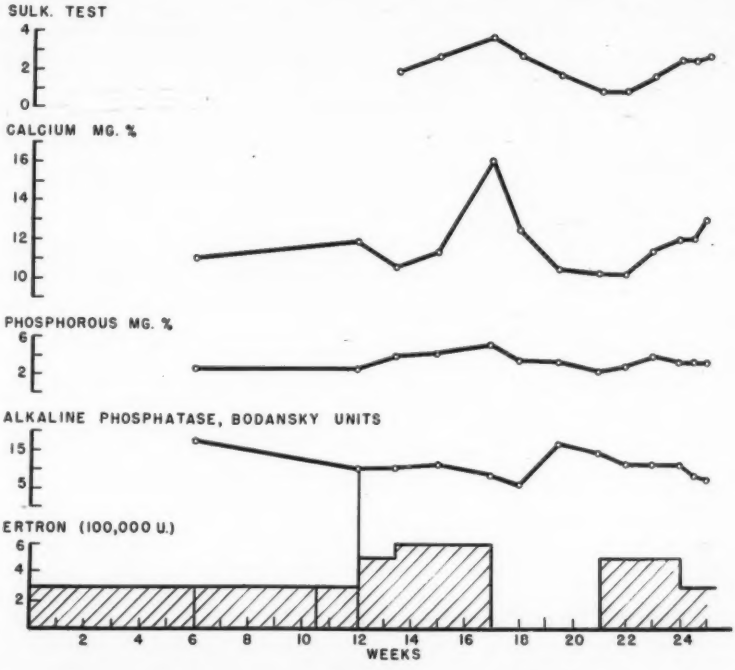

FIG. 5-F

Vitamin-D therapy and chemical response in patient S. L.

be prevented or controlled by early adequate vitamin therapy? Both of these problems can only be answered by a long follow-up. Since the failure to grow normally is assumed to depend upon the epiphyseal disturbance which is corrected by vitamin D, normal growth should follow. A definite statement regarding this point cannot, however, be made at this time, since systematic measurements have not been continued for a sufficient period of time. What evidence has been collected suggests that a normal growth pattern may be established. Three mothers have been especially impressed and have volunteered the information that, for the first time, the abnormal children seemed to be growing at a rate comparable with that of the rest of the family.

It has been stated that the metabolic error of this disease resolves at the time of epiphyseal closure. Albright[2] felt that in adults it becomes a chemical osteomalacia. Since most of the adults in this group have had recurrent deformities after epiphyseal closure, have a low serum phosphorous, and in some instances have Looser's lines, they probably need treatment. In others, no active disturbance was noted. The need for treatment in adults may depend on the severity of the initial disturbance.

SUMMARY

Vitamin-resistant rickets has been established as ordinary rickets which does not respond until massive doses of vitamin D are given. This study presents a series of twenty-five children and nine of ten involved parents who have been so classified. Accurate classification and diagnosis can be made by urinalysis and a few blood-chemistry studies. The most important of these are the determinations of serum calcium, serum phosphorous, and serum alkaline phosphatase. It is now felt that the disease is far more common than has been previously reported and actually represents the most common cause of dwarfism encountered in this Clinic. It has a strong familial tendency. It may be present in varying degrees of severity and the exact amount of vitamin therapy required for each individual case can only be determined by a trial-and-error method. It becomes less severe after the age of epiphyseal closure, but some adults may require continued vitamin therapy.

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DISCUSSION

Dr. G. Edmund Haggart, Boston, Massachusetts: The authors review the clinical findings presented by the syndrome of vitamin-resistant rickets in twenty-five children and nine of ten involved parents. They

point out that not only is this clinical picture more frequently seen than has heretofore been reported, but also that the disease has a strong familial tendency. Furthermore, this resistant rickets represents the most common cause of dwarfism they have encountered.

These observations are important since, with these facts in mind, osteotomies on severely deformed lower extremities in children will properly be withheld until an adequate study determines the presence or absence of vitamin-resistant rickets. If this deficiency is evident, then adequate therapy must be instituted before surgery is performed.

The authors agree that treatment is based on the threshold concept, as developed by Albright, and hence separate determinations for threshold level and the level of toxicity must be made for each patient. We support the belief that there is a relatively narrow margin between these two levels; and, therefore, in deciding on the vitamin-D dosage, particular reliance is placed on a determination of the blood-serum calcium, phosphorus, and alkaline phosphatase level, and the urinary calcium excretion, together with a study of the roentgenograms. Once on a maintenance dosage, the blood and urine studies should be made at a minimum of every two months.

That the authors are well aware of the dangers of the combination of massive doses of vitamin D in patients immobilized in plaster is evident by their report of seven patients, all of whom, while revealing marked improvement in the epiphyses, as shown by roentgenograms, developed symptoms of toxicity. I wish to ask if, in these patients, they were able to control toxic symptoms by simple discontinuation of the vitamin therapy and forcing fluids or were other measures also required.

Also, I am interested to learn if the authors have noted a hypercalcaemia in healthy patients, without evidence of vitamin deficiency, who have been immobilized in plaster following trauma.

When a long-term follow-up has been obtained, I hope the authors will report again on the question as to whether dwarfism can be prevented and the deformities controlled by early adequate vitamin therapy in these patients. Their present observations would certainly seem to indicate that a normal bone-growth pattern may be expected under appropriate treatment.

In conclusion this is a worth-while study, and I wish to congratulate the authors.

DR. WILLIAM T. GREEN, BOSTON, MASSACHUSETTS: We have had a very considerable experience with vitamin-resistant rickets over quite as long a period as that indicated by Dr. Pedersen. For one thing, we have now followed patients for a sufficiently long time to have been able to see them grow into maturity well controlled by adequate dosage of vitamin D. In those patients with marked deficiency, short stature approaching dwarfism is still present.

Resistant rickets is a disease that is roentgenographically and histologically typical of ordinary vitamin-D rickets, but very massive amounts of vitamin D are required for its control. There may well be other metabolic factors entering into its etiology. Certainly, it is a different problem from that of ordinary rickets.

I should like to emphasize certain features in the practical handling of these patients. We found that, if patients who were being treated with an adequate dosage of vitamin D were subjected to osteotomy, bothersome sequelae occurred in certain of them,—namely, hypercalcaemia and hypercalcuria. In certain patients, immediately after operation, there developed very high levels of calcium in the blood with a sufficiently high concentration of calcium in the urine to interfere with kidney function. It became routine, therefore, to discontinue treatment with vitamin D prior to the operation, to observe calcium in the blood and the urine at frequent intervals immediately postoperatively, and to resume the dosage of vitamin D as indicated. Frequently, the full dosage was resumed two weeks after operation.

Another important consideration is the fact that the dividing line in the dosage of vitamin D, which is adequate to control the rickets and that which may cause calcuria and kidney damage, is not very great. It is important, therefore, when these patients are first seen, to make frequent observations at the start in order to find the practical dosage of vitamin D. These observations should include appropriate blood and urine determinations. In practice, one of the important observations on the urine is a Sulkowitch test, which gives a rough, qualitative evaluation of calcium excretion in the urine. The mother of the child may be taught to do this determination, as it is very simple. Ordinarily, the urine by this test should show a very slight cloud; if the cloud is very dense, there is an overdosage of vitamin D, and if the urine is very clear, there is a possibility that the dosage is too little. The test is an aid in determining the dosage of vitamin D and in keeping the patient out of difficulty.

Vitamin-resistant rickets is most variable in its severity. There are patients who require just slightly more vitamin D than usual, and there are those who require several hundred thousand units a day. A low value of inorganic phosphorus in the blood serum is the rule in these patients, and in many of them the phosphorous remains quite low, even when the roentgenograms and other signs show healing. Recurrences of rachitic activity and deformities are apt to occur. Too frequently the story is that the activity of the process is controlled by adequate dosage of vitamin D, while the deformities are corrected by surgery. Then the patient disappears from observation; adequate vitamin-D therapy is not given; and the deformities recur. Adequate doses of vitamin D must be given until the patient's growth is complete.

I enjoyed this paper. It presents a thorough study of a subject with which we should all be familiar.

DR. M. BECKETT HOWORTH, NEW YORK, N. Y.: I had the privilege of a preview of this paper, at least in part, by seeing some of these patients in St. Louis several weeks ago. This group of patients with resistant rickets is the best worked up and most carefully studied with which I am familiar, and I can only confirm the observations the authors have made.

I have also had the privilege of seeing a number of patients with resistant rickets at the Babies' Hospital in New York, with Dr. Donovan McClure, who has been greatly interested in this subject. I would like to confirm the statement that the diagnosis is commonly missed. Nearly every patient whom Dr. McClure showed me had been seen and treated by an orthopaedic surgeon. They had had osteotomies—some of them had had two or three or four sets of osteotomies—without the diagnosis of vitamin-resistant rickets having been made. In every instance the deformities recurred. In some instances, the deformities were greater than they had been in the first place. I well recall female twins who had reached the age of twenty and still had evidence of active rickets in the epiphyses of the iliac crests as shown in the roentgenograms. The legs were so markedly bowed that these girls could hardly have stopped a pig between their legs. The rickets was controlled by large doses of vitamin D, and osteotomies were done with good permanent results.

It is difficult to say, at the moment, just how long before the operation vitamin-D therapy should be started, during what period it should be stopped, how soon it should be resumed, and how long continued after the operation. It is quite obvious that it is important that the rickets be kept under control, and the failure of osteotomy to correct permanently the deformity in these cases has often been due to failure to keep the rickets under control. Relapse has been common, and relapse has occurred in all the cases that we have seen, either because of neglect on the part of the patient or lack of knowledge on the part of the doctor. Now, perhaps, one word of caution might be justified. The curative dose in this disease is different from the maintenance dose; it might be that, if we continue giving the curative dose after the disease is arrested, instead of dropping down to the maintenance dose, harm may be done. Vitamin-D poisoning does occur. This perhaps has been the authors' difficulty with excessive doses of vitamin D. We must then be careful to keep the dose under control in order to avoid excess as well as to give enough.

I would like to raise one other point, the term "vitamin-resistant rickets". Perhaps we may be misled by using that term. We really do not know that this is a vitamin-resistant rickets. Perhaps there is another factor, or other factors, which may be responsible; and the mere fact that we have to give an enormous dose of vitamin D to cure this disease, or to keep it under control, does not necessarily mean that it is vitamin-resistant. Perhaps the other term which has been used, "refractory rickets", is less definitive and at the present moment of our knowledge, of more value.

DR. MCCARROLL (closing): With reference to toxic symptoms, in every instance in which toxicity has occurred, these symptoms have disappeared very rapidly within a period of ten days after withdrawal of vitamin therapy and the forcing of fluids. In one instance, there was a mild hematuria which also disappeared within a few days after treatment was discontinued. Regarding the incidence of hypercalcaemia in normal children while immobilized, we cannot answer this because we have not studied the problem thoroughly. We have seen children with other malformations of epiphyseal structures in infancy, with normal blood chemistry, in whom we used massive doses of vitamin D experimentally and in these children toxicity invariably developed very rapidly. In other words, children with vitamin-resistant rickets tolerate massive doses of vitamin D, whereas normal children will not tolerate it and will show evidence of toxicity within a few days.

The terminology which is applied to this disease has been questioned by Dr. Howorth. The term "vitamin-resistant rickets" has been used in this presentation because it has already become established and, at the moment, we know of nothing better.

We have presented this discussion for two reasons: First of all, to emphasize the marked frequency with which this disease occurs. It has been stated to be very rare and only thirty cases have been reported in the medical literature as far as we know. Yet in this series, twenty-five children and ten adults have been presented who we believe have been definitely proved to fall in this category. We first learned of this disease three years ago at an Academy meeting from W. T. Green. I had never heard of the disease at that time and apparently many others have also found this to represent a new field. I felt that we had cases in our Clinic that fell in this category and after returning home, this study was instituted. In years past, these cases have often been classified as dyschondroplasia, chondrodysplasia, and achondroplasia. We are thoroughly convinced that many clinics for crippled children in this country have children of this type classified incorrectly, who could be correctly classified with adequate study. The only studies needed are a complete urine examination with a Sulkowitch test and the three blood-chemistry determinations of calcium, phosphorous, and alkaline phosphatase. The second reason for presenting this study is to show, we believe very conclusively, the marked familial tendency which this disease presents. Of these twenty-five children, sixteen fell into ten family groups and in each of these ten families, at least one parent was involved.

We have seen only one infant who falls into this category, a seventeen-month-old baby of one of our ten family groups. This child has a low phosphorous, an elevated alkaline phosphatase, and some changes at the epiphyseal lines, which are considered indicative of this disease process. Unfortunately, this child had

(*Continued on page 224*)

SAFETY FACTORS IN THE USE OF THE PNEUMATIC TOURNIQUET FOR HEMOSTASIS IN SURGERY OF THE HAND*

BY JULIAN M. BRUNER, M.D., DES MOINES, IOWA

The pneumatic tourniquet, as employed and described by Harvey Cushing in 1904, has literally made possible modern surgery of the hand. It virtually eliminated the danger of nerve palsy, which often followed the use of the ordinary tourniquet. Cushing wrote:

"Surgeons and neurologists alike are familiar with the postoperative palsies of the extremities consequent upon the too energetic and prolonged application of an elastic tourniquet. Out of a considerable number of pressure paralyses, which have come under the writer's observation during the past two years, eight of them have thus originated. The greater number of these were of the brachial type, the tourniquet having been employed to render bloodless the field at the elbow usually for resections of the joint. A thin, flabby arm, especially in these cases of joint disease and in spite of all precautions, lends itself readily to such an injury as the poorly protected nerves are easily compressed against the hard shaft of the humerus . . . .

"The suggestion of an inflatable tourniquet doubtless originated in the use of the distensible armlet of the Riva-Rocci blood-pressure apparatus. On one or two occasions I had utilized the apparatus itself as a means of shutting off the circulation during cocaine operations on the hand, by holding the pressure slightly above that of the registered systolic arterial pressure. Objections are self-evident. Although the pressure of the armlet is relatively painless, its slow distension, by shutting off the veins before the arteries, causes such an engorgement as to render a clean dissection most difficult. Consequently a similar armlet, though broader, of less distensible rubber and of such quality that it would stand boiling, was constructed, and by connecting it with a bicycle pump of sufficient size one or two quick strokes of the piston sufficed to fill it. Such an arrangement I have made use of many times for cocaine operations on the extremities with the greatest satisfaction . . ." [5].

The employment of any tight constriction about the arm, such as a tourniquet, may be locally harmful in two ways: first, by injury to those structures beneath it through direct pressure, as in the case of the peripheral nerves; second, by injury to those tissues distal to the tourniquet as a result of congestion or ischaemia. Although instances of nerve injury are now rare**, there is clinical evidence that prolonged tourniquet application may not be entirely without harm to the tissues of the forearm and hand under certain circumstances.

Complete arrest of the circulation in the upper extremity is obviously unphysiological. Yet the assertion is frequently made that such stoppage may be continued for periods of one and one-half to three hours without harm. We doubt that this is true. It has not as yet been shown clinically or experimentally just how long surgical ischaemia may be safely prolonged, what individual variation there is in tolerance to ischaemia, or exactly what factors play a part.

Because of widespread use of the pneumatic tourniquet in surgery of the hand, and of its potential dangers, this matter is of great importance. The literature is almost devoid of specific information on this subject. We believe that prolonged application of the pneumatic tourniquet for surgery of the hand may have adverse effects upon the tissues of the hand, the extent of which depends upon several factors.

Bunnell has stated[4] that after excessive periods of tourniquet ischaemia, there is some degree of "tissue reaction". Presumably such a reaction is the result of biochemical changes in the tissues associated with ischaemia, cell damage incidental to anoxia, and nature's efforts to repair such damage.

Instances of this sort have been seen occasionally after periods of tourniquet applica-

* Read at the Annual Meeting of the American Society for Surgery of the Hand, February 10, 1950, New York, N. Y.

** In 1945, a case of nerve palsy due to pneumatic-tourniquet application was observed in an army hospital. Paralysis involved muscles supplied by the radial, ulnar, and median nerves. Recovery gradually took place after several months. This untoward result was probably due to prolonged excessive pressure in the arm cuff (around 500 millimeters of mercury) caused by an inaccurate aneroid gauge.

tion, ordinarily considered safe, in patients whose tissues apparently have a low tolerance to ischaemia. The effects are usually first noticed at the time of the first postoperative dressing. The manifestations observed are as follows:

1. Puffiness of the hand and fingers (not a pitting oedema) evidenced by a smoothing out of the normal skin creases;

2. Stiffness of the hand and finger joints to a degree not otherwise explained;

3. Color changes in the hand, which is pale in the horizontal position, more so when elevated, and congested in the dependent position;

4. Subject sensations of numbness in the affected hand without true anaesthesia;

5. Objective evidence of weakness in the muscles of the forearm and hand without real paralysis.

Such a puffy, pale, stiff hand has been seen in cases in which the factors of operative trauma, or postoperative immobilization of the hand, were inadequate to explain these phenomena, and in which hematoma formation and infection had not been present. We believe that ischaemia during surgery is the chief cause of such a condition; therefore, the condition might be called "post-ischaemic hand syndrome".

This syndrome is not identical with Volkmann's ischaemic palsy, which is supposedly produced by a different mechanism,—namely, prolonged ischaemia associated with congestion. To simulate the conditions causing Volkmann's palsy, one should maintain a tension in the tourniquet somewhere between systolic and diastolic blood pressure; whereas, in this case a pressure considerably above systolic blood pressure has been maintained with resulting complete ischaemia.

What, then, are the conditions which may combine to produce an untoward result as a consequence of tourniquet ischaemia? While it is only truthful to say that the exact factors are unknown, the following deductions may be made from clinical and experimental evidence:

Age Factor: Young individuals seem to show a high degree of what may be called ischaemia or tourniquet tolerance. In children, we have never observed the syndrome. In military service, soldiers in the younger age group showed great tolerance in this respect, unless their hands had suffered previous extensive vascular damage or scarring. In civilian practice, we have occasionally noted in middle-aged individuals, apparently in good health, the tendency for a puffy, pale, stiff hand to develop postoperatively; in elderly patients the tendency is greater.

Stiffening Factor: In adults there is an additional factor which does not run parallel with the age factor, but nevertheless bears some relation to it. This may be called the "stiffening factor", for want of a better term, and to avoid using the terms "arthritic" or "rheumatic". It is well known that stiff joints have a much greater tendency to develop in some individuals than in others, following immobilization, trauma, *et cetera.* It is probable that such persons also have a low tolerance to other tissue insults, such as ischaemia, especially in regard to postoperative joint mobility.

Time Factor: After short operations requiring the use of the tourniquet for twenty to thirty minutes, untoward tissue reaction has not been seen. Operations of fifty to sixty minutes' duration enter the time zone of possible danger in certain individuals with low tourniquet (ischaemia) tolerance. In operations of one to two hours, which usually entail extensive dissections of the hand or forearm, definite precautions must be taken to prevent tissue damage from ischaemia, including properly spaced periods of tourniquet release for adequate "breathing periods".

General Factors: It is obvious that patients with advanced vascular disease, rheumatoid arthritis, *et cetera*, are poor prospects for surgical ischaemia.

Local Factors: Local vascular damage, as a result of fresh trauma, or poor blood supply, as a result of old cicatrix, may make use of the tourniquet hazardous.

Temperature Factor: It has been shown experimentally by Allen and others that

tourniquet ischaemia may be maintained for long periods if the temperature of the tissues is kept low, thus keeping metabolism at low ebb. The application of a tourniquet to the leg of a laboratory animal is safe for many hours in an ice bath, but, if the tissues are warm, gangrene may result. Since relatively minor degrees of tissue damage in the human subject cause concern, the reasons for avoiding overheating of ischaemic tissues on the operating table are all the more compelling.

The principle of safeguarding ischaemic tissues by cooling, following arterial occlusion by embolism, has been generally accepted. Should not the same principle be applied to tissues under surgical ischaemia, which are suddenly deprived of blood supply more completely than by any embolus?

What other steps can be taken to minimize possible harm from tourniquet ischaemia in surgery of the hand? The proper selection of patients for elective hand surgery with respect to age, stiffening factor, and general and local vascular integrity will do much to prevent harm.

Careful choice of the operation to suit the patient is of equal importance. It is of doubtful value to relieve a moderate Dupuytren contracture in the hand of an elderly patient, if, following the operation, his hand does not regain its normal flexibility. Prolonged tourniquet ischaemia is undoubtedly one of the factors which may produce this result. Individuals with known stiffening tendencies should not be subjected to long application of the blood-pressure cuff.

Tourniquet time should be shortened as much as possible. This may be accomplished in several ways: by careful preoperative planning of the work to be done, by rapid dissection, by elimination of waste motions, and by performing certain phases of the operation without benefit of ischaemia when this does not slow up operative procedures. This is often possible in the closing stages of an operation, after active bleeding has been controlled, and oozing has stopped.

Recovery from tissue damage in the hand due to ischaemia is variable. In the young, this occurs rapidly and completely, while in older persons resolution may be slow. In occasional cases, residual effects—namely stiffness and puffiness—may persist for several months. In rare instances, late fibrotic changes may take place, manifested by induration throughout the soft tissues of the hand and permanent stiffening of joints. These effects have been ascribed to tourniquet ischaemia only when all other factors could, in fairness, be eliminated.

Specific rules for tourniquet application are not generally agreed upon, but we consider the following ten points to be important:

1. The only safe tourniquet is the blood-pressure cuff attached to a mercury manometer. The rubber bag in the cuff and its connections must be absolutely leak-proof, for a pinhole will allow the pressure to fall gradually below systolic level, resulting in congestion, which, if prolonged, is more dangerous than ischaemia.

2. The upper arm should first be padded with several turns of sheet wadding, after which the manometer cuff is snugly and smoothly applied and held in place by wide roller bandage.

3. Before inflation of the cuff, the hand and forearm should be emptied of blood by elevation for several minutes, or should be stripped of blood by means of an Esmarch bandage (except in the presence of infection).

4. The pressure in the cuff must be applied very quickly, as noted by Cushing, to prevent blood from entering the forearm while the cuff is being inflated.

5. Pressures of 270 to 300 millimeters of mercury are used for adults; 250 millimeters or less for children.

6. Time limits are not agreed upon. We believe that one hour of ischaemia is safe for healthy adults below middle age; in older patients, caution should be used.

7. Re-application of pressure after a ten-minute period of release will be necessary

in many cases to complete operative work. Time limits for the second or third periods of ischaemia are unknown. A possible summation of tissue reaction must be taken into account.

8. Before the relase of tourniquet pressure, the open surgical wounds of the hand or forearm should be covered with saline compresses and snugly bandaged; the arm should be elevated. Unless this is done, the wave of arterial blood which surges back into ischaemic tissues on release of the tourniquet will result in great tissue congestion. Pressure on the open wounds should be maintained for five to ten minutes to allow cessation of capillary oozing, after which bleeders may be caught.

9. Colored skin antiseptics should be avoided for they may be misleading. The ruddy tint of antiseptic paint may impart to the skin a sanguine glow, when actually the tissues are in a state of suspended animation. The unpainted skin gives valuable information to the surgeon relative to ischaemia, cyanosis, or congestion due to a fall in tourniquet pressure.

10. During the period of ischaemia, the tissues distal to the tourniquet should be kept cool. Hot spotlights must not be used. On the basis of all experimental work, frequent irrigation of the tissues in the operative field with cool or cold saline is indicated.

In conclusion, we feel that the use of tourniquet hemostasis in surgery of the hand should be regarded not as a necessity, but as a luxury, to be used sparingly and intelligently. Every tissue has its limit in regard to ischaemia and the hand is no exception.

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DISCUSSION

Vitamin-Resistant Rickets

(*Continued from page 220*)

not been followed long enough to be included in the present study, but it is with the young infants that the greatest good may be accomplished. If seen very early in life and if adequate treatment is instituted, it will be possible to prove beyond doubt whether or not adequate continued vitamin therapy will bring about spontaneous improvement in the deformities and thus alleviate the necessity for surgery. It will also be possible to determine whether or not the obvious dwarfism can be controlled. These questions cannot be answered from our present study. At least three of these families, however, have stated that the involved children, under adequate vitamin therapy, have shown a rate of growth which is comparable to that of the normal children in the family, for the first time in their lives.

SCOLIOSIS WITH PARAPLEGIA

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Paralysis of the lower extremities is an infrequent complication of scoliosis. In the author's examination of many hundreds of scoliotic patients during the past forty years, only two cases have been seen. The first was reported in 1923[2]; the second will be recorded here. McKenzie and Dewar, in a recent review of the literature, collected forty-one cases, including five of their own. To this number may be added two patients recently reported by Love and Erb and one by Hyndman, making a total of forty-four cases. When one contemplates the many thousands of scoliotic patients seen by orthopaedic surgeons, one appreciates that the incidence of paraplegia is, fortunately, very small.

The case to be described presents unusual clinical features which made an early accurate diagnosis difficult; it emphasizes the importance of early laminectomy and the variety of technical detail which may be required for adequate decompression of the cord.

A. B., a Puerto Rican girl, twelve years of age, was admitted to the author's service on May 26, 1947, with the chief complaints of inability to walk and increasing deformity of the back. This girl had apparently been well until the age of ten years. At that time, she fell while roller-skating and thereafter suffered from pain in the mid-portion of her back. Three months later, she noticed increasing difficulty in walking; the family then sought medical advice. The locomotor problem was attributed to her obesity, for which thyroid was prescribed. She did not improve, and shortly thereafter she was admitted to a hospital in San Juan, where she remained for two years. A diagnosis of Pott's disease was made, with paralysis of the lower limbs, and she was treated by continuous recumbency, either on a rigid bed or on a convex frame. Since improvement did not follow, the parents brought her to the Hospital for Joint Diseases.

The first examination revealed a very obese girl, apparently in good general condition, not suffering any pain, able to sit up, but unable to stand or walk. The upper extremities, chest, and abdomen were normal except for several pigmented areas in the skin of the abdomen. No abnormal masses were visible or palpable anywhere in the body. The heart and lungs were normal and the blood pressure was 110 systolic, 70 diastolic.

The lower extremities lay completely extended at the hips and knees, with the feet in equinus of about 120 degrees. There was bilateral patellar and ankle clonus; the Babinski sign was present bilaterally. There was some power, although evidently greatly subnormal, in the muscles about the hips and knees. There was some power of contraction in the calf muscles, but no power in the anterior and lateral leg muscles on either side. There was a defect in the sensory responses to the level of the eighth thoracic dermatome.

The back presented a marked kyphos at the thoracolumbar junction with flattening of the thoracic area and an increased lumbar hollow. There was no tenderness of the spine and no evidence of fluctuant masses anywhere in the back.

The head and face were normal. She had well developed sexual characteristics. There was no adenopathy. The cranial nerves functioned well, and the pupils reacted to light and accommodation. The blood and urine showed normal findings; the Kahn and Kline tests were negative. The sedimentation rate varied from 45 millimeters per hour at the time of admission to 7 millimeters just before operation.

Roentgenograms showed a marked deformity of the spine at the thoracolumbar junction, which was very unusual in appearance (Fig. 1). The thoracic spine was in the mid-line down to and including the tenth thoracic vertebra; the ninth and tenth thoracic vertebrae were irregular and somewhat cuneiform in outline, the two vertebrae forming a poorly outlined quadrilateral mass. A wedge-shaped hemivertebra was interposed on the right side, between the tenth and eleventh thoracic vertebrae. Distal to this point the spine turned sharply to the left, almost at a right angle. The curve involved the eleventh and twelfth thoracic and the two proximal lumbar vertebrae, below which the lumbar vertebrae gradually returned to the mid-line over the sacrum. The left thoracolumbar curve was short and the vertebral elements were ill-defined. The X-ray Department defined the curvature as "angular kyphoscoliosis", which was, indeed, a very accurate word picture of the deformity. The basis of the scoliosis was apparently a supernumerary hemivertebra on the right side between the tenth and eleventh thoracic vertebrae. For a more accurate study of the osseous pathology, laminographic anteroposterior roentgenograms were made. These confirmed the previous interpretation by disclosing: "The triangular supernumerary body is situated to the right side of the spine between the tenth and eleventh dorsal segments. There is no evidence of any recent infectious process."

Since there was evident compression of the cord, myelographic studies were instituted, pantopaque

being used. The fluoroscopic and roentgenographic examinations of the neural canal were reported as showing complete arrest of the column (of pantopaque) at the level of the eleventh thoracic vertebra. Roentgenograms of the chest showed no evidence of tuberculosis of the lungs. It was finally concluded that the patient had paraplegia, due to an osseous deformity of the spine, a congenital kyphoscoliosis with compression of the spinal cord.

The differential diagnosis was reviewed, since some features of the deformity were susceptible of various interpretations. The following diagnoses were considered:

1. *Tuberculosis:* This patient had been treated for Pott's disease. She came from a country where tuberculosis was prevalent. There was considerable haziness and loss of definition of the bones at the thoracolumbar junction, findings in favor of a chronic lytic process. Against the diagnosis of tuberculosis were: (a) the absence of roentgenographic evidence of lung tuberculosis; (b) the absence of a paravertebral or other "cold" abscess; (c) the lack of roentgenographic signs of an inflammatory or unequivocally destructive process; and (d) the absence, clinically, of local tenderness to pressure over the thoracolumbar kyphos.

2. *Von Recklinghausen's Disease:* The one positive finding consisted of pigmented spots on the abdomen. There were no neurofibromatous masses or cystic disease of any of the bones. We were aware of the report of a case of von Recklinghausen's disease with scoliosis and paraplegia [4], but our patient presented no additional confirmatory evidence of this diagnosis. Incidentally, none has appeared during the two years subsequent to the laminectomy.

3. *Injury:* The possibility was entertained that the injury at the age of ten years had initiated a scoliosis secondary to inordinate compression of the vertebrae at the thoracolumbar junction. Against this diagnosis was the fact that symptoms of cord compression did not appear until three months after the accident. Had the trauma caused a crushing fracture-dislocation of the spine, the cord symptoms would undoubtedly have appeared promptly.

4. *Congenital Scoliosis:* The presence of a unilateral wedge-shaped mass between the tenth and eleventh thoracic vertebrae was the strongest evidence in favor of congenital scoliosis due to a hemivertebra. It was recalled that, in most of the reported cases of scoliosis with paraplegia, the etiological basis was a congenital osseous defect. What was perplexing in this case was the angular sharpness of the curvature, together with the indistinctness of the vertebral bodies immediately above and below the hemivertebra. Fortunately, tomographic studies were available; they confirmed the presence of a hemivertebra and the absence of any inflammatory process.

The most logical diagnosis was considered to be congenital scoliosis with paraplegia, and it was decided that a decompression laminectomy should be performed.

On September 2, 1947, bilateral laminectomy was done on the area from the ninth thoracic to the second lumbar vertebrae. The cord did not pulsate. There was no granulation tissue to indicate the existence of inflammation or trauma. There appeared to be a definite constriction in the region of the twelfth thoracic and the first lumbar as a result of the spine deformity; when this constriction had been removed, there was observed to be easy passage of a probe distal to the area of laminectomy. The dura was not opened. The wound was closed in layers and the patient left the operating room in good condition.

The operating surgeon did not, unfortunately, specify the nature of the constriction, except to indicate that it was the result of the spine deformity. We may assume that the constriction consisted of a reduction in the capacity of the neural canal. It is regrettable that the dura was not opened since, in the majority of the reported cases which have been improved by the operation, the dura was opened and the cord was liberated at the apex of the deformity for a distance of several inches.

Following the laminectomy there was slow, partial improvement. Several weeks after operation the patient was able to flex and extend her toes. By March 1949, one and one-half years after the decompression laminectomy, she had regained sufficient strength in the muscles about the hips and knees so that she was able to stand and walk with the aid of crutches. (Fig. 2). Sensation had improved somewhat. By November 1949, sensation in both lower limbs was almost normal. She had recovered enough power in all the muscles about the hips and knees so that she could elevate the limbs from the bed, although she could not keep

them raised for more than a minute or two. Thus the laminectomy resulted in only partial improvement.

When this patient was examined in March 1949, the muscles about the hips and knees were improved and there was no deformity at any of these joints. However, both feet were still in marked equinus; there was no voluntary dorsal flexion on the right side and very little on the left. In an effort to produce further improvement in the patient's ability to stand and walk, a posterior bone block and subtalar and calcaneocuboid arthrodeses were performed on the right with excellent results. Six months later the right foot was in excellent relation to the leg and in only slight equinus. The ankle clonus which had been present prior to the stabilization was absent. A few months after the operation on the right, a stabilization operation was performed on the left foot. Since the muscles of the back and the hip extensors were fairly strong, it was felt that this patient might ultimately be able to stand and walk without crutches.

PATHOGENESIS OF PARAPLEGIA COMPLICATING SCOLIOSIS

The direct and immediate cause of the cord compression and paralysis is not certain. Several causes have been identified, and there may be others. Chief among these are the following:

1. *Unequal Growth of Dura and Spinal Column*

McKenzie and Dewar believed the most frequent cause of unequal growth to be a

FIG. 1

FIG. 2

Fig. 1: Anteroposterior view of spine shows thoracolumbar angular scoliosis with a congenital hemivertebra interposed on the right side between the tenth and eleventh thoracic vertebrae.

Fig. 2: After operation patient was able to stand with aid of crutches. Note severe kyphoscoliosis.

tight *dural canal.* This opinion was apparently based on the fact that the greatest improvement following laminectomy occurred in those cases in which the dura had been incised and left open. The basis for the tight dura is the assumed unequal growth of the dura and spinal column. McKenzie and Dewar stated ". . . the exciting cause which precipitates paralysis is the rapid growth of the spinal column together with the inability of a tight dura to accommodate itself to such growth". This, of course, may be true; it appears speculative and improbable, however, because growth, unless influenced by some selective factor, intrinsic or extrinsic, involves all tissues. A more likely cause for the paralysis is:

2. *Sudden Increase in Scoliosis*

In thirty-five of the forty-one cases reported by McKenzie and Dewar, the paralysis appeared during the period of rapid growth, at a time when the scoliosis, if not controlled by treatment, is subject to a sudden and marked increase. Almost all of the reported cases are of the kyphoscoliotic variety, in which a marked increase in severity occurs during the second period of rapid growth. McKenzie and Dewar cited a case in which a known sudden increase in the deformity was followed by paralysis. The patient was a child who had begun to show signs of neurological involvement at about the age of six years. The scoliosis was treated and the symptoms were arrested. At the age of ten, the scoliosis suddenly increased, the symptoms returned, and in a few weeks there was complete paraplegia.

Love and Erb described a boy of eleven, who had scoliosis secondary to an attack of poliomyelitis at the age of two years. When seen in 1945, he had some neurological symptoms. Orthopaedic treatment, including spine fusion, controlled the symptoms for two years, after which there was a sudden appearance of complete spastic paralysis. Here, too, the aggravation occurred during a period when the scoliosis is subject to a rapid increase.

It is logical to assume that the tightness of the dura, causing the paralysis, results from an increase in the scoliotic deformity to which the dura has not, in these exceptional cases, accommodated itself.

3. *Anomalous Tight Dural Band*

McKenzie and Dewar reported that in one of their cases there was found, at operation, a linear thickening of the dura which extended obliquely across the neural canal and compressed the cord.

4. *Spur of Bone in Neural Canal*

A spur of bone along the neural ring can cause pressure on the spinal cord in scoliosis, as it can in many other osseous lesions of the spinal column. McKenzie and Dewar reported such a case.

5. *Angulation of the Cord*

In the face of the extensive architectural and morphological changes in the vertebrae, it is a matter of great surprise that there is not more frequent involvement of the spinal cord, especially in some of the paralytic scolioses with marked lateral distortion. One must assume that severe angulation and kinking of the cord would, at times, cause an interruption of nerve impulses and produce paralysis. Such a condition was found by Love and Erb in their second case of scoliosis. It should be emphasized that the level of the cord compression always corresponds with the apex of the curvature—that is, the site of the greatest deviation and rotation deformity of the spine—and hence the site of the greatest potential angulation of the cord. In Hyndman's case: "It was concluded that the angulation of the cord had been exaggerated rather acutely, and that this was the factor responsible for loss of function".

6. *Tightness of Nerve Roots*

In a number of the patients operated upon, especially in recent years, the nerve roots at the level of the cord compression were unusually taut; mobilization of the dura and freedom of the cord were established only after the tight nerve roots had been severed. Whether this is a primary or secondary lesion is difficult to state, but the observers have emphasized the importance of severing all tight nerve roots. Fortunately, no consequential damage occurs at a level requiring such surgery.

7. *Injury*

The relation between trauma and paralysis in scoliosis lies in the potential aggravation of the spine curvature following an injury. The history in many cases of scoliosis indicates that the deformity increases after an injury. Trauma — not only a sudden accident but also laborious occupation, as in Hyndman's case—induces stretching of the tissues, similar to that occurring during mobilization by gymnastic exercises, which allows an increase in the deformity. In the case reported here, there was an unequivocal history of injury, after which neurological symptoms appeared, and the deformity was recognized. In Love and Erb's second scoliosis case (their Case 5), the neurological symptoms appeared after a fall in a hockey game. The dura was thickened and adherent to the arachnoid, suggesting trauma with subsequent organization. Trauma, therefore, is a factor, although as yet of undetermined extent, in the aggravation of scoliosis.

From the preceding considerations, we may summarize the pathological findings in paraplegia complicating structural scoliosis as consisting of one or more of the following changes at the apex of the deformity: (1) a tight dura; (2) an anomalous dural band; (3) bony spurs; (4) post-traumatic inflammatory tissue; and (5) angulation of the cord.

TREATMENT

The treatment naturally falls into two categories, conservative and operative. McKenzie and Dewar reported that, of twenty-four patients treated conservatively, eleven improved; yet they concluded that laminectomy is advisable and should not be delayed too long. In view of the pathological changes at the site of compression of the cord, one can hardly hope for complete relief from paralysis through any non-surgical method except possibly at the inception of neurological symptoms. At that time one could conceivably relieve the pressure on the cord by reducing the scoliotic deformity, fusing the spine, and protecting it against relapse of the curvature by providing a supporting apparatus to be worn continuously until after puberty, when there would be little likelihood of an increase in the deformity. Under all other circumstances, and surely in the presence of a paralysis which is not quickly relieved by conservative treatment, has appeared suddenly, is marked, or is advancing rapidly, surgery is indicated without delay. Now that surgery is relatively safe, the treatment of choice is immediate laminectomy. Delay subjects the patient to irreversible damage to the spinal cord.

Laminectomy is performed to decompress the spinal cord. The details of the operation depend upon the pathological changes found when the dura and cord have been exposed. One must bear in mind the variety of pathological changes already encountered, and be alert to additional alterations not yet appreciated or described. In the past, a variable portion of one lamina or the whole of several laminae on the concave side of the curve were removed. In the light of recent experience, one must look for anomalous soft-tissue formation, bone spurs, and unusual angulation of the cord; and be prepared not only to enlarge the neural canal and open the dura, but to remove any crests of bone anterior to the cord or to perform a liberal costotransversectomy for anterior or lateral transplantation of the cord to a degree that will permit adequate correction of the pathological angulation.

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ADOLESCENT DEFICIENCY DISEASE

REPORT OF A CASE

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The classical variety of rickets seen in infants is gradually disappearing from clinics in the United States, but is being replaced by several interesting variations. These variations are usually seen in older children and adolescents, and are well suited for clinical, chemical, and physiological study. It is hoped that a sufficient number of cases will be studied to provide additional knowledge, particularly in regard to excretion and absorption of vitamin D and to calcium and phosphorus metabolism.

For a review of the literature, the reader is referred to the excellent paper by McCune, Mason, and Clarke. Similar cases have been reported by Albright, Butler, and Bloomberg; Eliot and Park; Bakwin, Bodansky, and Schorr; and Brakeley.

J. T., a white girl, aged thirteen, was first seen on February 24, 1947, as an ambulatory patient; she was admitted to the New Jersey Orthopaedic Hospital on February 28. For two months this child had complained of painful feet, and her gait had been peculiar. She had noticed increasing ease of fatigue, but thought she needed new arch supports. She would walk home from school (a distance of six blocks) and then rest the remainder of the day. Any activity that required walking was shunned because it tired her. The parents had noticed the gradual appearance of a peculiar waddling gait. Marked effort was required when the child arose from a chair or climbed stairs; these functions were carried out slowly and with great difficulty. The symptoms gradually grew worse and, about two weeks prior to examination, there developed severe aching pain in the left thigh; she was barely able to walk or to move her legs.

The child's birth had apparently been normal and uneventful, but the mother thought there was a disproportion between the legs and the trunk,—either the legs were unusually thin or the trunk was unusually large. Her obstetrician saw nothing unusual in the infant's appearance. Later the baby "filled out beautifully". Her development was normal up to the age of eight months.

Starting at three months, the child had received one teaspoonful of cod-liver oil and ten drops of viosterol in fresh orange juice daily; the orange juice was increased gradually to eight ounces daily. Her appetite was good; the bowels always functioned normally. She was fed by breast and bottle until the age of three months, and given a proprietary fortified milk thereafter. She never crawled, but at eight months she tried to stand. She cried in apparent pain whenever she tried to stand, and any pressure on the feet caused crying. A pediatrician made a diagnosis of rickets. Medication consisted of six teaspoonfuls of cod-liver oil and fifty drops of viosterol daily, and a "powdered form of calcium". The mother was told not to permit the child to walk, and the diet was ordered to include beef juice, liver, fish, home-made soup, and "pastina". This regimen was continued for two years without interruption.

The child was allowed to walk at eighteen months, wearing special shoes. Her development in all respects was then normal until the present illness. The teeth erupted on time, the first one at the age of five months, and she had no more than three cavities. About one year before admission, the patient complained of painful feet when walking and standing; a chiropodist prescribed arch supports and special shoes, with

apparent relief. For two years a rash had been present in the antecubital and popliteal fossae, which left the skin thickened, rough, and scaly; it would bleed easily when scratched. The condition appeared to be a psoriatic lesion. It was worse in summer. The mother thought the child was sensitive to fresh fruit and tomato juice.

The child's habits and intellectual development had been normal. She was socially well adjusted. Her menses began at the age of eleven and were on a normal twenty-eight-day cycle, lasting five days.

The family history showed that the father and mother and both maternal and paternal grandparents were living and well. There were no familial or hereditary diseases. As far back as can be traced by the parents, there had been no intermarriage. The patient's brother, aged nineteen, and one sister, aged fifteen, were living and well. Neither sibling has had a similar illness.

Physical examination showed a fairly well developed and somewhat obese young white girl, not appearing either acutely or chronically ill. There were no gross deformities. She walked with a peculiar waddle, suggestive of that seen in cases of muscular dystrophy. On the right, the waddle or limp was somewhat gluteal in character and the buttocks were unusually prominent. The general physical examination was negative, and skeletal and muscle examination was essentially negative from the waist up. All the muscles about both hip joints showed weakness, especially the flexors. Both quadriceps were weak, probably more so on the right. Both thighs and calves showed generalized muscular atrophy, but the muscles of the feet were fairly strong and well developed. Moderate bilateral genu valgum was noted, and there was second-degree pes planus on the left with some hallux valgus and a very prominent navicular. The right foot showed first-degree pes planus without hallux valgus, and the navicular was less prominent. Examination of the hips showed no telescoping; the Trendelenburg test was negative. There was very slight limitation of internal rotation of the hip bilaterally. Otherwise, all hip, knee, and ankle motions were normal. Reflexes were normal; no sensory changes were noted.

Roentgenograms on February 24 were reported as showing definite fuzzy demineralization of the bones of the entire pelvis, lower lumbar spine, sacrum, hips, and upper ends of the femora. The iliac crests were indistinct; the epiphysis could not be seen. There appeared to have been complete obliteration of the sacro-iliac joints (Fig. 1). The rami of the pubis and ischium were very irregular in outline and showed spotty demineralization, giving a moth-eaten appearance. Just distal to the capital femoral epiphyseal plates were broad bands of demineralization. The cortices of the pelvic bones appeared unusually thin. Anteroposterior views of both shoulders, chest, and thoracic spine showed generalized bone demineralization, and there were broad bands of demineralization in the metaphysis next to the proximal humeral epiphyseal plates. The humeral cortices were unusually thin. The tenth rib on the left side showed a cystlike area of demineralization near its spinal articulation. Lateral views of the lumbosacral spine and sacrum showed such marked demineralization that the spines were barely visible and the bodies were very hazy in outline. Anteroposterior views of the wrists, knees, and ankles showed as the outstanding feature a broad band of demineraliza-

FIG. 1

Pelvis and hips on admission, showing rachitic changes especially about the sacro-iliac joints.

FIG. 3

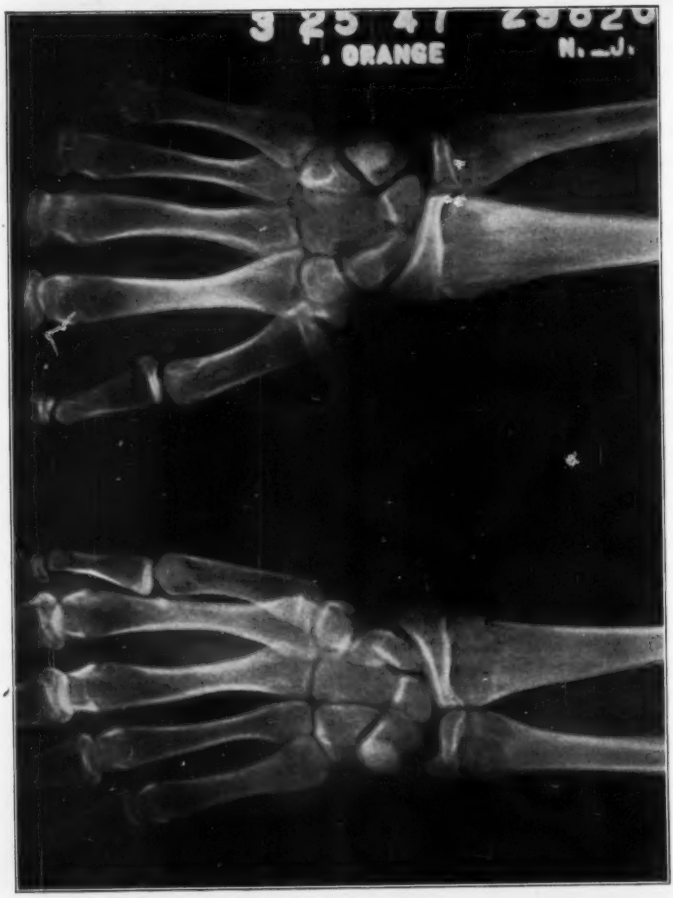

FIG. 2

Fig. 2: The wrist joints on March 25, showing the rachitic changes.
Fig. 3: The right wrist after healing.

FIG. 4

Pelvis and hips after healing. Note the reappearance of distinct sacro-iliac joint lines. The appearance of new bone along the iliac crest may possibly be suggestive of hypervitaminosis.

tion just proximal to the epiphyseal plates of both radius and ulna. There were coarse trabeculations in this area, parallel to the long axis of the bone. The epiphyses themselves looked fairly normal except for slight demineralization. Soft-tissue swelling was not noted.

Laboratory findings on admission were as follows:

Red blood cells	4,250,000
Hemoglobin	86 per cent.
White blood cells	6,800
Polymorphonuclear neutrophils	68 per cent.
Lymphocytes	30 per cent.
Monocytes	1 per cent.
Eosinophils	1 per cent.

The serum calcium was 10 milligrams per 100 cubic centimeters, serum phosphorus 2 milligrams per 100 cubic centimeters; the serum alkaline phosphatase was 22 Bodansky units. The spinal fluid showed total serum protein of 29 milligrams per 100 cubic centimeters; the cell count was 3. The Wassermann reaction and colloidal-gold curve were negative. The blood sedimentation rate was 8 millimeters per hour (Cutler method). Urinalysis was entirely negative except for a trace of albumin and an occasional white blood cell. The non-protein nitrogen was 33 milligrams and the blood creatinine was 1.5 milligrams per 100 cubic centimeters. Determination of the serum alkaline phosphatase was repeated four days later and showed 26 Bodansky units.

The patient was admitted to the New Jersey Orthopaedic Hospital for further study. Her general course became rapidly worse; within a few weeks she was unable to walk and was barely able to lift herself onto a bedpan. She was quite irritable. However, her appetite continued to be fairly good. On March 12, under general anaesthesia, a small block of the left ilium and sacro-iliac joint was removed for study. At operation it was noted that the bone was very soft and unusually vascular. Hot wet packs were required to control the oozing. Her recovery from the operation was uneventful.

The tissue obtained was studied by Henry L. Jaffe at the Hospital for Joint Diseases, New York City. His report was as follows:

"The fragments of bone submitted for examination showed, in the tissue sections, widening of the proliferating cartilage zone and the presence, beneath the cartilage zone, of rather thick trabeculae with wide osteoid borders. Altogether, the cytological tissue changes are those of rickets or are ricket-like.

"The roentgenograms reveal widening of the metaphyseal regions and a wide, proliferating zone immediately beyond the cartilage plates.

"The blood-chemistry findings (sugar 81 milligrams, non-protein nitrogen 22 milligrams, calcium 9.8 milligrams, inorganic phosphorus 3.0 milligrams per 100 cubic centimeters, and alkaline phosphatase 41.3) are in line with a rachitic or rachitoid state.

"One can eliminate renal disease (renal rickets), primary hyperparathyroidism, and by the history, idiopathic steatorrhoea. The question narrows down to whether this is rickets caused by vitamin-D deficiency or endogenous rickets of the Fanconi syndrome type. The disease picture as a whole tends to favor the latter, but the inorganic phosphorus, although low, is not strikingly so. If it is endogenous rickets, the phosphorus will fall further in the next year or so."

Dr. Jaffe also suggested the possibility of failure of absorption of vitamin D from the gastro-intestinal tract. With this thought in mind, the patient was started on an intramuscular dose totaling 3,100,000 units of vitamin D per week, as well as 210,000 units of vitamin A, 3,150 milligrams of vitamin C, 126 milligrams of vitamin B_1, 84 milligrams of vitamin B_2 (riboflavin), 12.6 milligrams of vitamin B_6 (pyridoxine), 630 milligrams of niacinamide, 42 milligrams of calcium pantothenate, and 168 milligrams of mixed tocopherols. She was permitted to receive treatment at home. The response was dramatic and rapid, and muscle power began to return. First she was able to use the bathroom; she gradually became able to walk longer distances, felt better in general, had complete relief from pain, and improved in her disposition.

On May 1, her blood calcium was 11.2 milligrams and blood inorganic phosphorus 4.2 milligrams per 100 cubic centimeters; the alkaline phosphatase was 15.8 Bodansky units. On June 4, her blood non-protein nitrogen was 41 milligrams, blood creatinine 1.5 milligrams, blood calcium 10.0 milligrams, and inorganic phosphorus 4 milligrams per 100 cubic centimeters; the alkaline phosphatase was 10.2 Bodansky units. In fear of hypervitaminosis, the intramuscular vitamin D was discontinued and the vitamin-D capsules were reduced to one, three times daily. This decreased the dosage of vitamin D to a daily intake of 150,000 units. On June 17, her blood non-protein nitrogen was reported as 20 milligrams, blood calcium 10.8 milligrams, and blood inorganic phosphorus 3.8 milligrams per 100 cubic centimeters; the alkaline phosphatase was 2.5 Bodansky units.

By July 9, when the next blood-chemistry determination was done, the patient was completely symptom-free and was leading a perfectly normal life. (A few weeks previously, she had been able to take part in her graduation exercises at school.) Her non-protein nitrogen was reported as 21.4 milligrams, calcium 12 milligrams, and phosphorus 3.6 milligrams per 100 cubic centimeters; the alkaline phosphatase was 6.4 Bodansky units.

Anteroposterior roentgenograms of the right wrist (Fig. 3), right ankle, and right knee, taken on July 16, showed almost complete calcification of the previously noted band of rarefaction, just proximal to the epiphyseal plates of the joints. The epiphyses of the knee and ankle had almost closed. An anteroposterior view of the pelvis, as compared to previous New Jersey Orthopaedic Hospital films, showed increased calcific density of the bones; for the first time, the sacro-iliac joints could be defined (Fig. 4).

On July 16, the dosage of vitamin D was reduced to one capsule per day. Her clinical recovery seemed complete and her activities were those of a normal child of her age. On August 27, her blood non-protein nitrogen was reported as 31 milligrams, blood calcium 10.8 milligrams, and blood inorganic phosphorus 4.1 milligrams per 100 cubic centimeters; the alkaline phosphatase was 7 Bodansky units. On November 5, 1947, her blood non-protein nitrogen was 40 milligrams, calcium 10.0 milligrams, and phosphorus 3.5 milligrams per 100 cubic centimeters; the alkaline phosphatase was 7 Bodansky units.

At this time, because of the continued normal reports and the maintenance of clinical recovery, the medication was discontinued. The patient has remained completely symptom-free and normal in all respects.

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TRAUMATIC PARALYSIS OF THE SERRATUS ANTERIOR RELIEVED BY TRANSPLANTATION OF THE RHOMBOIDEI*

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Various operations to correct weakness or paralysis of the shoulder have been suggested. Fixation of the scapula by connecting fascial strips to the spine was described by Armitage Whitman. Transplantation of a portion of the pectoralis major by a fascial attachment to the medial border of the scapula has been recommended by Tubby, Durman, Dickson, and others [3,4].

In a recent case of traumatic paralysis of the shoulder, when conservative measures such as physical therapy and support failed to relieve the winging and loss of elevation after six months, a relatively simple procedure was tried which proved very satisfactory. The rhomboidei and the trapezius were used.

CASE REPORT

B. M., a male, aged twenty-seven, was a passenger in an automobile on June 25, 1948, and was asleep when a collision occurred which killed the driver. When examined at the Physician's Hospital, Warrenton, Virginia, the patient was in acute distress, complaining of discomfort in breathing and of pain in the cervical region and in the left index finger. There was a bruise over the left anterior shoulder region, marked deformity of the left index finger, and two large lacerations of the face. The neurological findings were that the motor system and cranial nerves were intact; the extremities moved without limitation.

The following day the patient was removed to the George Washington University Hospital, where he came under the author's care. A dislocation at the proximal interphalangeal joint of the index finger was reduced, the wounds were dressed, the lacerations were sutured, and, after five days, the patient left the Hospital. Two days later he complained of numbness and weakness of the right upper extremity. Examination showed limited ability to abduct the extremity; an area of anaesthesia, 9 by 4 centimeters, could be outlined on the lateral surface of the arm. The deltoid seemed very flabby and, at the origin near the acromion, a sulcus could be felt. Because the posterior portion of the muscle was detached from the bone, a repair was carried out. An abduction brace was applied for several weeks and, after the muscle had healed, movement of the shoulder was satisfactory to about 90 degrees. However, marked winging of the scapula was seen and the vertebral border was so far away from the chest wall that the examining hand could palpate the anterior surface (Fig. 1). When the scapula was held firmly, the arm could be elevated above shoulder level. Conservative treatment was tried for six months, with very little improvement. Thereafter the operative procedure to be described was carried out, with an excellent result.

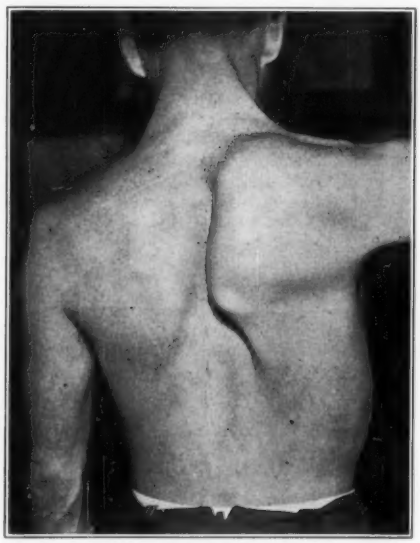

FIG. 1

Showing the limitation of abduction and marked winging of scapula.

Operative Technique

With the patient lying prone, an incision was made along the entire medial border of the scapula. The trapezius was retracted medially (Fig. 2). The bone was stripped with a periosteal elevator, the muscle tissue being kept in a continuous sheet. The stripping was continued on the anterior surface for about one inch along the entire length of the vertebral border. While the scapular border was brought as close to the spine as possible, the muscle sheet, consisting of the rhomboideus major and some of the fibers of the rhomboideus

* Presented at the meeting of the Washington Orthopaedic Club, January 1950.

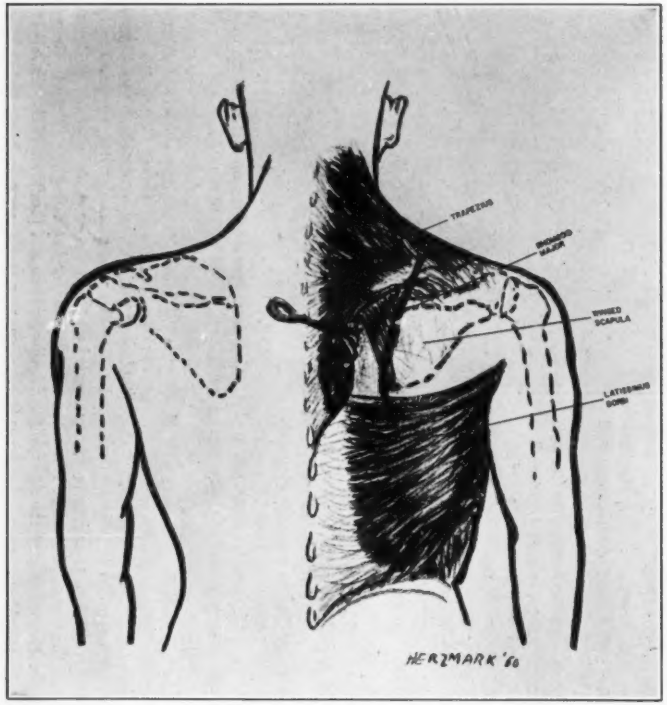

FIG. 2

Black line shows course of incision. Trapezius is retracted medially. Rhomboidei shown attached to border of scapula.

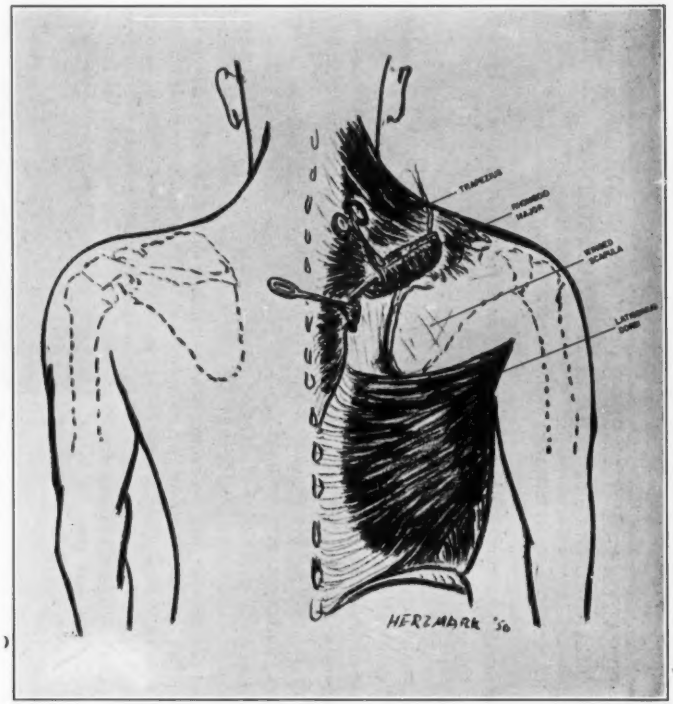

FIG. 3

Scapular attachment of rhomboideus major and rhomboideus minor has been dissected free.

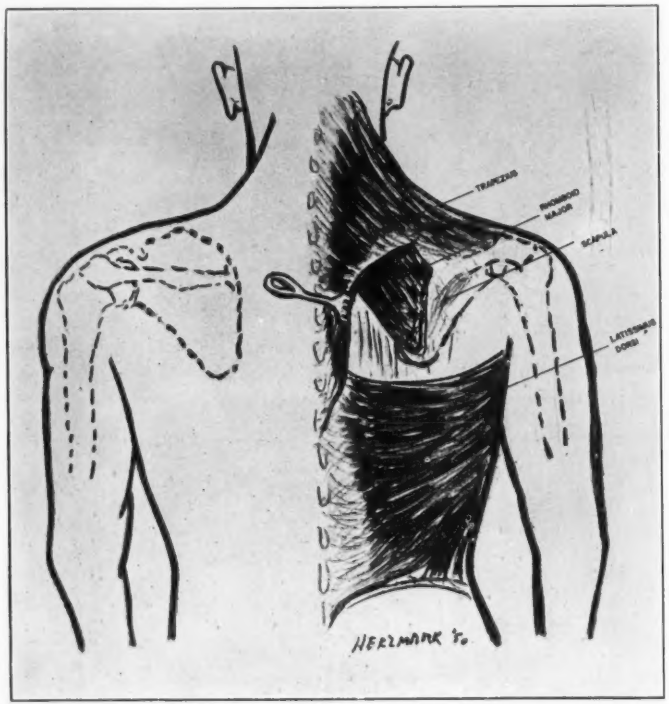

FIG. 4

Rhomboideus major and minor have been transplanted to posterior surface of scapula. Scapula is drawn as far medially as possible.

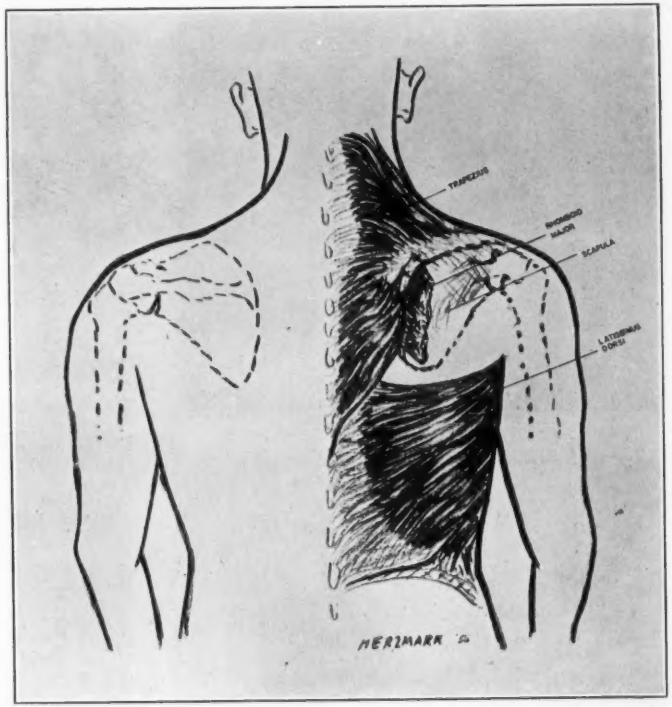

FIG. 5

Trapezius has been replaced and sutures have been inserted in fascial portion over rhomboidei.

FIG. 6-A

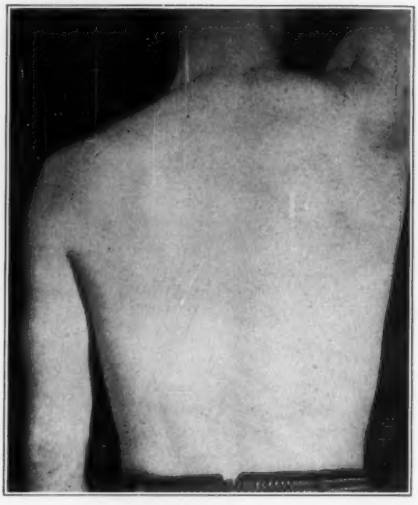

FIG. 6-B

Appearance one year after operation. There has been complete restoration of function.

minor, was reattached posteriorly, and as far laterally as it would stretch without too much tension, to the infraspinatous fascia (Figs. 3 and 4). The tendinous border of the trapezius was then sutured over this and the skin was closed (Fig. 5). A Velpeau type of bandage was applied.

After four weeks the bandage was removed, and mild physical therapy and gentle exercises were used to build up the strength of the transplanted muscles. At the end of three months complete elevation was possible without any "winging" of the scapula. A recent examination, a year after the operation, showed normal action of the shoulder girdle and satisfactory strength of the muscles (Figs. 6-A and 6-B).

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AN UNUSUAL CASE OF PAGET'S DISEASE

Presenting an Extraordinary Degree of Osteoblastic Activity *

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There is considerable evidence that the etiology of Paget's disease has its basis in a vascular disorder. Osteitis deformans was considered by Paget to be an inflammatory disease of the skeleton, but he did not emphasize the increased vascularity. Edholm, Howarth, and McMichael have since noted that the blood flow through the bones involved in Paget's disease was increased up to twenty times of normal.

Generalized osteitis fibrosa cystica was considered by von Recklinghausen to be related to Paget's disease, if it were not the same disease. He did note, however, that excessive bone formation was a characteristic of Paget's cases, while his cases showed predominantly cystic changes in bone. Schmorl, in describing the histological mosaic pattern of Paget's disease, laid the groundwork for the differentiation between the two diseases.

FIG. 1-A

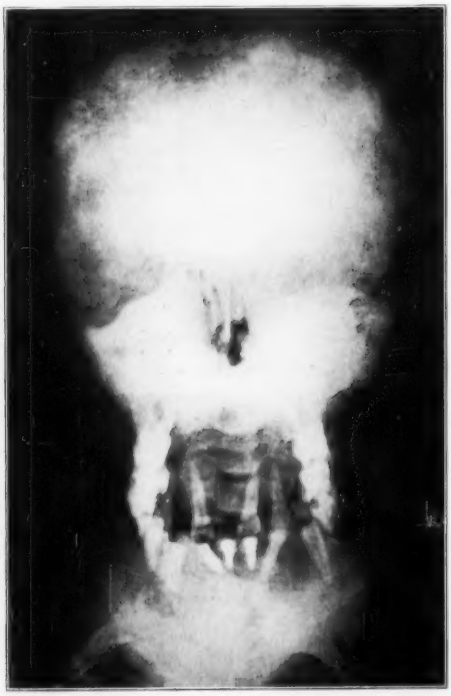

FIG. 1-B

Fig. 1-A: Photograph, October 1947, shows enlargement of the head, the maxilla and the mandible. Atrophy of the upper extremities and "clubbing" of the fingers should be noticed.

Fig. 1-B: X-ray, October 1947, demonstrates typical "cotton wool" appearance of the skull, enlargement of the maxillae and the mandible, wide separation of the teeth, and the absence of the lamina dura.

* Based on Scientific Exhibit at the Annual Meeting of The American Academy of Orthopaedic Surgeons, New York, N. Y., February, 1950. (Exhibit was awarded medal for "originality".)

TABLE I

CHARACTERISTICS IN THIS CASE RESEMBLING VON RECKLINGHAUSEN'S DISEASE OR PAGET'S DISEASE OR BOTH

Characteristics	von Recklinghausen's Disease Osteitis Fibrosa Cystica Osteitis Fibrosa Generalisata	Paget's Disease Osteitis Deformans Bone Dystrophy	This Case
Generalized distribution	Yes	No	No
Parathyroid affected	Yes	No	Yes
Calcification of organs	Yes	Yes	Yes
Serum calcium	Raised	Normal or slightly raised	Normal 10.8
Serum phosphorus	Decreased	Normal	Normal 3.5
Serum alkaline phosphatase	Normal or increased	Increased	Increased 28.9
Serum protein	Normal	Normal	Normal 6.31
Decalcification of bone	Yes	Yes	Yes
Lamina dura present	No	?	No
Bone destruction (resorption)	Yes	Yes	Yes
Bone production	No or slight	Yes (marked)	Yes (marked)
Bone cysts (histological)	Yes	No	Yes
Tumors of supportive tissue	Yes	No	No
Line of demarcation between bone and disease	No	Yes	Yes
Deformities due to stress	Yes	Yes	Yes
Increased blood supply to bone	No	Yes (marked)	Yes (marked)
Architectural integrity maintained	Yes	No	No
Pain in bones	Yes	Yes	Marked
Skull thickened	No	Yes	Yes
Skull moth-eaten	Yes (finely)	Yes (coarsely)	Yes (coarsely)
Osteoblastic activity	No	Yes	Yes
Cement lines	Regular	Irregular	Irregular

Mandl reported marked improvement in cases of osteitis fibrosa cystica when parathyroid adenomata were removed. Hyperparathyroidism, rather than von Recklinghausen's disease, has become the accepted term for the disease which is primarily a metabolic disorder.

Although from the pathologist's viewpoint there still exists confusion between the two entities in borderline cases, Paget's disease is not a manifestation of hyperparathyroidism; the two have been shown to coexist in cases reported by Albright and Reifenstein.

CASE REPORT

The patient, a colored female, aged twenty-six years was admitted to the University Hospital on October 28, 1947. Her chief complaint was "hurting" in both legs. In 1943, at the onset of illness, she had noticed aching in both the thighs and the knees, which recurred every few months. In 1945, she had noticed a continuing enlargement of her facial bones, and during the years 1946 and 1947, she had been able to get around the house only by sliding in a chair. She had had five children, four of whom were dead. A hysterectomy had been performed at the time of the Caesarian delivery of the last dead infant in May 1946.

The physical examination showed a poorly nourished, colored female in no acute distress. Her blood pressure was 120 systolic and 75 diastolic; her temperature 99 degrees; her pulse, 110; and her respiration, 16. There was an obvious enlargement of the cranial and facial bones, and the mandible was extremely large and protruding. The nares had flattened and the septum and the bridge had almost disappeared. The alveolar processes had greatly increased in size with the oral mucosa stretched tightly over the bone. Lymph nodes were enlarged and non-tender. There was fixed kyphosis in the thoracic region, lordosis in the lumbosacral area, and scoliosis affecting the lumbar vertebrae; the thorax was barrel-shaped and emphysematous; the ribs were close together. The thoracic wall was pliable, and there was marked angulation of the costochondral junction. The lung fields were clear; and abdominal findings were negative. The pelvic girdle was abnormal in shape; and the extremities were small and atrophic.

Roentgenograms revealed a tremendous thickening of the bones of the skull and of the face. The collapse of the vertebrae, together with an irregularity of the cortices of the long bones with some cystic changes, was also shown.

The Kahn test was negative.

Blood chemistry findings were as follows:

Acid phosphatase, 0.31 Bodansky units,
Alkaline phosphatase, 28.9 Bodansky units,
Total protein, 6.31 grams per 100 cubic centimeters of serum (albumin, 3.48 and globulin, 2.93),
Calcium, 10.8 milligrams per 100 cubic centimeters of serum,
Phosphorus, inorganic, 3.5 milligrams per 100 cubic centimeters of serum,

FIG. 2-A

FIG. 2-B

Fig. 2-A: A profile photograph, October 1947, depicting the elongation of the jaws and the enlargement of the head. Note engorgement of the facial and the temporal vessels.

Fig. 2-B: X-ray, October 1947, shows that the changes of the skull correspond to the changes typical of Paget's disease. Particularly notable are the enlargement of the maxillae and the mandible. Note that the upper three cervical vertebrae appear unaffected.

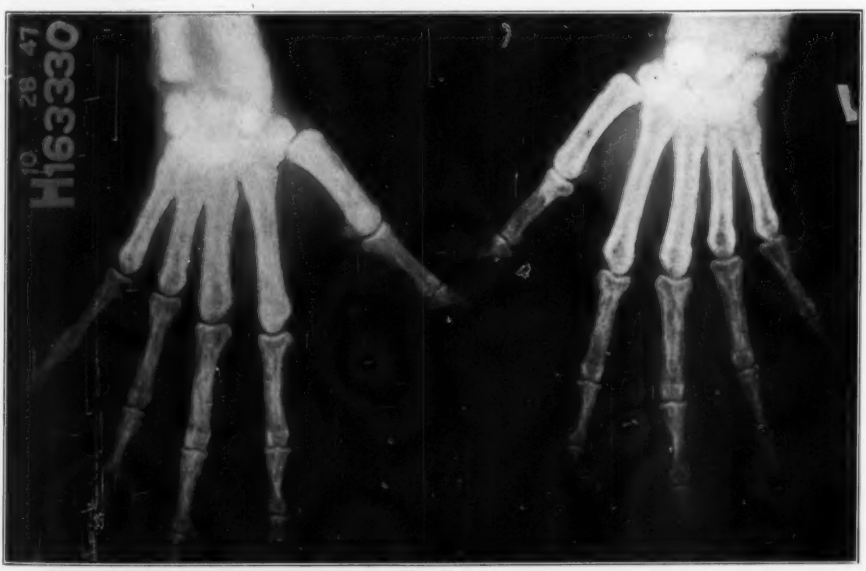

FIG. 3

Roentgenogram, October 1947, demonstrates the changes of structure of the metacarpals and the phalanges; note absorption and production, and the "clubbing" of the terminal phalanges.

FIG. 5

FIG. 4

Fig. 4: Roentgenogram, October 1947, of the femora, demonstrating the loss of density, absorption and production.
Fig. 5: Roentgenograms, October 1947, showing changes of the tibia and fibula, the radius, the ulna and the hand typical of Paget's disease.

FIG. 6-A

FIG. 6-B

Fig. 6-A: Photograph of autopsy specimen showing the tremendous overgrowth of the maxilla and the mandible.

Fig. 6-B: Roentgenogram showing skull changes and the marked bony overgrowth of maxillae and mandible. Note that upper three cervical vertebrae are apparently unaffected.

Non-protein nitrogen, 19 milligrams per 100 cubic centimeters of serum.

Urinanalysis revealed sugar 89. The test for Bence Jones protein was negative.

A biopsy of the left tibia on November 12, 1947, showed excessively active fibrosis of a few minute spicules of bone and an occasional fragment of osteoid tissue. This change is compatible with Paget's disease (Fig. 7-A).

The patient was treated symptomatically and later left the hospital against medical advice on November 14, 1947. Although she refused further hospitalization and study, contact was not entirely lost. She died in March 1949, and the body was obtained for autopsy and skeletal study.

Table I indicates how the condition in this patient resembles Paget's disease and von Recklinghausen's disease, as well as how some of the characteristics are indicative of both diseases.

AUTOPSY FINDINGS

Gross

The body was that of an adult, colored female, weighing approximately thirty-five kilograms. The frame work was extremely mis-shapen and the body measured approximately 87.5 centimeters in length. Emaciation was present.

There was an extreme enlargement of all the facial bones, especially of the maxillae and mandible; the oral cavity was almost completely occluded. Enlargement of the other facial bones had produced a flat contour to the face with the skin stretched tightly over the surface. The nose was flat, only the prominent nares suggesting evidence of the pre-existing structure.

The greatly enlarged and protruding mandible measured 20 by 18 by 15 centimeters and the measurement from the vertex of the skull to the tip of the mandible was 36 centimeters. A few widely spaced "peg teeth" projected from the smooth surface of the mandibular mass. The mouth was almost obliterated by enlargement of the maxillae, the palate, and the mandible; the oropharynx was similarly narrowed, barely admitting a finger from below.

The pupils were round and equal.

The scalp stripped easily. The calvarium measured 3.0 centimeters in thickness. The diploe and the tables of the calvarium were replaced by a pinkish-white, soft, gritty tissue which could be cut with a knife. The dura was extremely difficult to remove from the calvarium.

No noteworthy changes had taken place in the parenchyma of the brain which weighed 1,287 grams. The sella turcica was shallow, while the dorsum sellae turcicae and the clinoid processes were thickened. No noteworthy changes of the pituitary glands were found.

Many of the vertebral bodies were compressed and soft.

The upper and middle lobes of the right lung and the upper lobe of the left lung felt nodular to palpation. These nodular areas on cut section resisted the knife and had a woody character.

The pericardial cavity, heart, oesophagus, thyroid, and larynx showed no noteworthy changes. One of the right parathyroids was enlarged and measured 2.0 centimeters; two of the left parathyroids were enlarged and each measured 0.5 centimeter in diameter. The peritoneal cavity, liver, gall bladder, spleen, pancreas, adrenals, kidneys, bladder and urethra, genitalia, and gastro-intestinal tract showed no noteworthy changes.

Microscopic

Many foci of calcification were found in the walls of the vessels and in the interstitial tissues of the lungs.

There were vascular and peritubular calcifications of the kidneys and some foci of calcification in the glomeruli. There was calcification of the vessels of the pancreas; calcification of the vessels of the bladder; and calcification of the vessels of the skin. No calcification of the aorta was found, but there was calcification of

FIG. 7-A

Biopsy specimen of the tibia, October 1947. Photomicrograph (× 78) shows an area of active fibrosis in cancellous bone and decalcification of the pre-existing bone.

FIG. 7-B

Autopsy specimen, March 1949. Photomicrograph (× 78) shows new-bone formation encroaching upon the medulla. The cortical and cancellous bone have been largely replaced by poorly calcified, irregular trabeculae of osteoid tissue which are supported by cellular fibrous tissue.

the anterior tibial artery and vessels of like size. Hyperplasia of all the parathyroids was found as well as calcification of the vessels of the parathyroids.

FIG. 7-C

Autopsy specimen, March 1949. Photomicrograph (× 78) shows that the pre-existing bone has been replaced by poorly calcified trabeculae of osteoid tissue supported by cellular fibrous connective tissue. Osteoclastic absorption of the newly-formed osteoid tissue with replacement by cellular fibrous tissue should be noted. There is an abundance of osteoclasts.

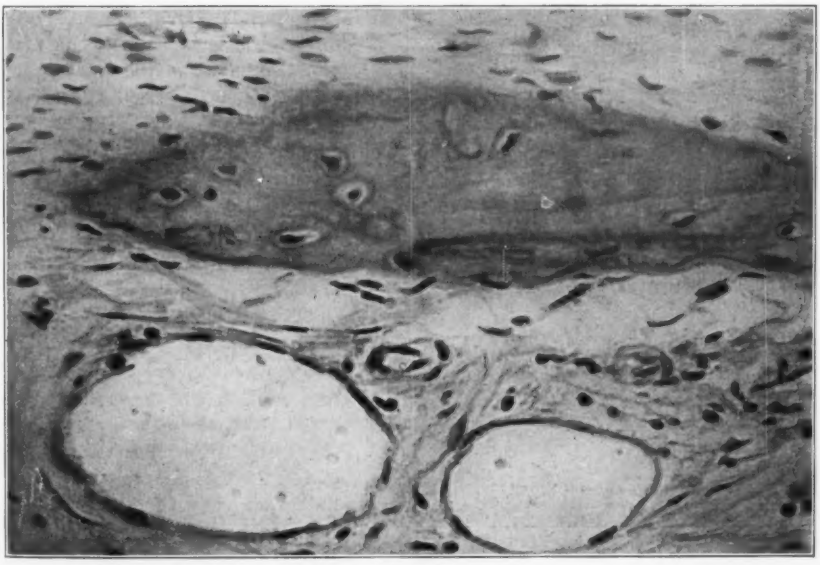

FIG. 7-D

Autopsy specimen, March 1949. Photomicrograph (× 345) shows highly magnified spicules of newly formed, poorly calcified osteoid tissue with some irregularity in the cement lines. Within the fibrous stroma, there are two microcysts.

Subperiosteal and cortical bone had been replaced or had thickened, while the diploe of the cranium had been obliterated. The marrow cavity of the long bones had narrowed and no pre-existing bone remained, having been replaced by poorly calcified, irregular trabeculae of osteoid tissue, which lay in cellular and acellular fibrous tissue. The older osteoid trabeculae had undergone osteoclastic absorption. Some areas were composed of cellular fibrous tissue and contained many osteoclasts which had absorbed the few remaining fragments of osteoid tissue. Microscopic cysts were prominent in many areas. The pattern of the cells in the bone structure resembled osteitis fibrosa.

COMMENTS

Under stress, diseased bones undergo great changes. This is more marked in the lower than in the upper extremities and more marked in the sacrum and lumbar spine than in the thoracic and cervical spine. It was noted that the upper three cervical vertebrae were not involved.

Immobilization favors decalcification, for it removes from the bones the stimulus of production. The marked resorption can be explained on the basis that the patient was bedridden for about two years prior to her death. However, the marked production in the mandible, maxillae, and the skull can be explained by the stress of the temporal muscles and that of the muscles used in eating and swallowing. The effort of the patient to eat and swallow must have been terrific, and we might expect proliferation to be in a direct ratio to the increased vascularity resulting from the effort. Figures 1 and 2 show the engorgement of the facial and temporal vessels.

Since deafness has been considered a frequent complication of Paget's disease and since this patient was not deaf even on the day of her death, we decided to remove the ossicles of one ear. These were found to be perfectly normal and unaffected by the disease. We concluded that the bone formed in cartilage probably escaped the disease process. The ossicles, derived from Meckel's cartilage, were therefore unaffected.

FIG. 8

Photograph of the interior of the skull base showing apparently unaffected area. This portion of the skull is formed in cartilage. The defect in right petrous bone is due to incision made for removal of the ossicles. Insert shows unaffected ossicles.

The base of her skull was little if at all involved in the disease process.

In reviewing the literature, it was noted that the deafness noted in 90 per cent. of the cases of Paget's disease was caused by bilateral chronic otitis media and the remaining 10 per cent. by other causes, including otosclerosis.

The question of leontiasis ossea naturally presented itself. Just what this term implies seems not to be clear and, since three types are recognized, —the Virchow, the von Recklinghausen, and the Paget—it would be natural to assume leontiasis ossea to be the proliferation of the bony elements of the face. It is presumed by some that the first two groups, Virchow and von Recklinghausen, are identical with fibrous dysplasia.

Syphilis was ruled out in this case.

Melorheostosis, thought by some be a descriptive term and not a disease

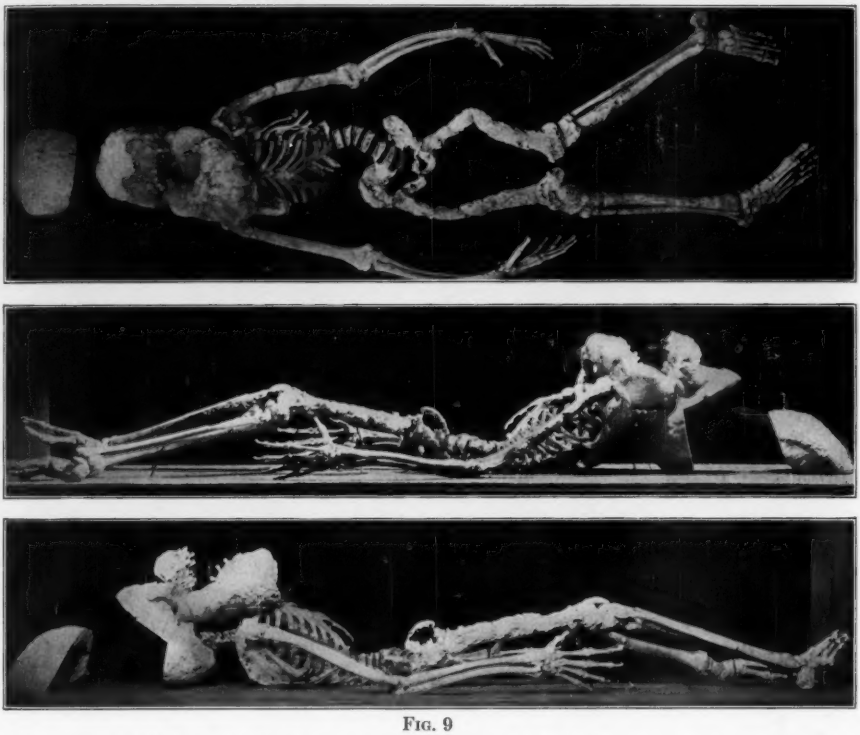

FIG. 9

Photograph of skeleton with skull cap removed, illustrating skeletal changes and affording a comparison of the amount of involvement in the various regions.

entity, is used to describe marked proliferation even of the facial bones and mandible. Either the terms leontiasis ossea or melorheostosis can apply in this case of Paget's disease, which possesses some concomitant findings which are so typical of von Recklinghausen's disease. The conclusion is that at least these two diseases are present in this case.

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CONGENITAL ABSENCE OF THE SACRUM AND COCCYX

Report of Two Cases

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Congenital absence of the sacrum and coccyx, known as sacrococcygeal agenesis, is a rare anomaly. In a review of the literature [3-5], the authors have found forty-five reported cases of partial or complete absence of the sacrum and coccyx. To this number, two cases are added:

Case 1. R. A. D., a four-year-old white girl, was admitted to the Cooper Hospital for observation because of a congenital defect of the lower extremities, and for bladder and bowel incontinence.

The father had congenital club feet. The mother's pregnancy had been normal in every respect. One older sibling was normal.

The child had had numerous orthopaedic devices applied to the lower extremities, including bilateral casts and Denis-Browne splints. The parents had been told that she was being treated for "spina bifida".

Physical examination showed the upper portion of the torso to be fairly well developed, but there was marked failure of development from the hips down. Atrophy of the buttocks, thighs, and calf muscles was most apparent. The child was able to stand with assistance, with the lower extremities in universal flexion. Examination of the spine revealed a bone defect between the ilia. Rectal examination showed absence of the bony concavity of the sacrococcyx. Both hips appeared to be normal. Both feet were semirigid, the right being held in mild equinus, the left in equinovarus.

Roentgenograms of the lumbosacral spine (Figs. 1-C and 1-D) revealed an absence of the fifth lumbar body and of the entire sacrum and coccyx. The right and left pelvic girdles were closely approximated in the mid-line. A survey of the remaining skeleton disclosed no further abnormalities.

Case 2. R. V., a one-year-old white boy, was admitted to the Children's Ward, Cooper Hospital, for

Fig. 1-A

Fig. 1-B

Case 1. Photographs of child at age of four years.

orthopaedic treatment. The diagnosis of sacrococcygeal agenesis had been made by our orthopaedic surgeon at another hospital when he had first seen this infant three days after birth (Fig. 2-C).

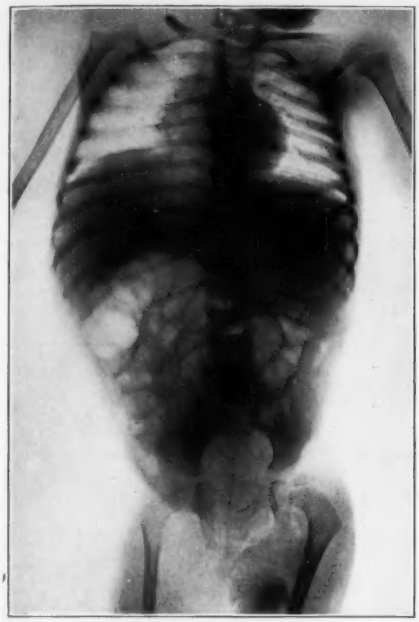

FIG. 1-C

FIG. 1-D

Rotentgenograms of spine showing absence of fifth lumbar body and of entire sacrum and coccyx.

FIG. 2-A

FIG. 2-B

Case 2. Photographs taken when infant was one year of age.

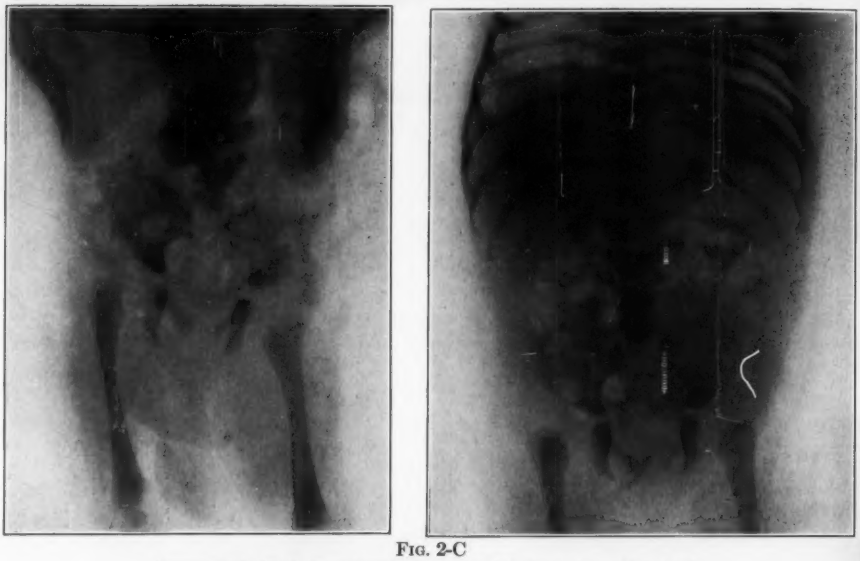

FIG. 2-C

January 26, 1948. Roentgenograms of spine three days after birth.

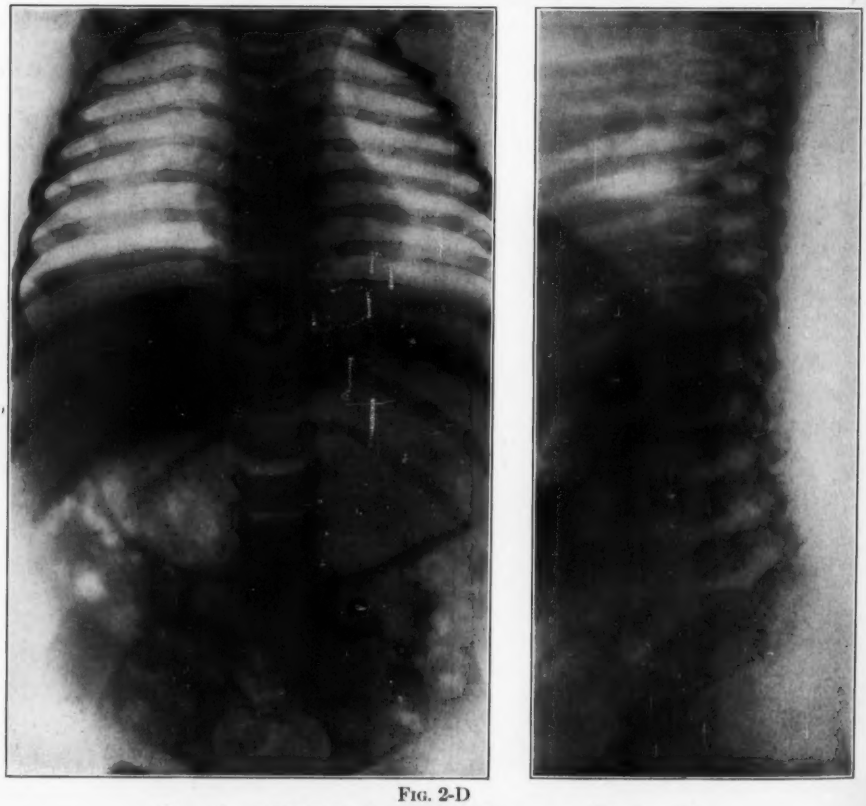

FIG. 2-D

November 6, 1948. Roentgenograms taken at age of nine months.

FIG. 2-E

November 9, 1948. Lateral view of lumbosacral spine.

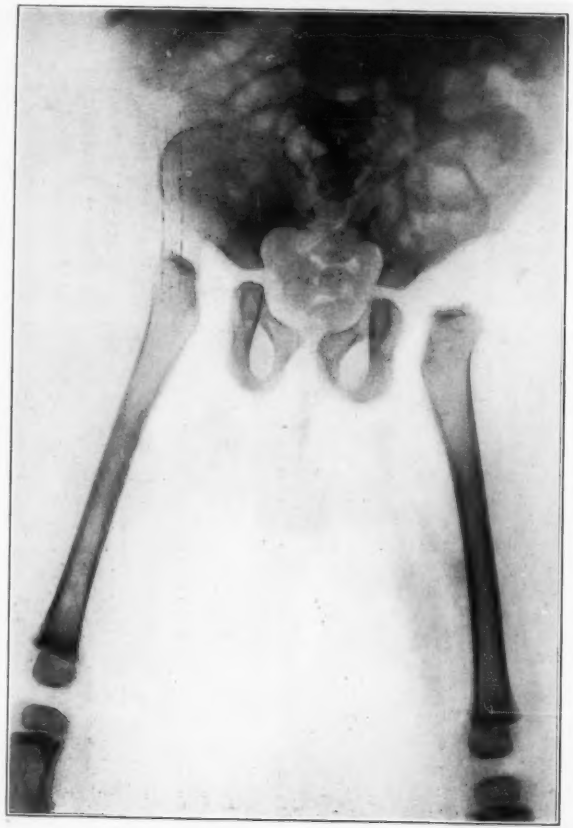

FIG. 2-F

February 18, 1949. Anteroposterior and lateral views when child was one year of age.

The family history was negative for congenital or skeletal defects, except for bowing of the tibia in one first cousin. The mother's pregnancy had been normal. As in Case 1, one older sibling was perfectly normal.

Physical examination at the age of three days had revealed a prominence over the lumbar area. Both hips were dislocated and were in marked flexion. The legs were rotated on the thighs. Both feet showed severe talipes equinovarus.

Roentgenograms of the lumbosacral spine (Figs. 2-D to 2-G) showed complete absence of the sacrum and coccyx, with deformity of the fifth lumbar vertebra. Further roentgenographic examination of the osseous system revealed fusion of the second and third ribs, and bilateral dislocation of the hip.

DISCUSSION

These two cases appeared on the Children's Ward at the same time. The features manifested by each were almost identical, the difference in development being due to the age variation of three years.

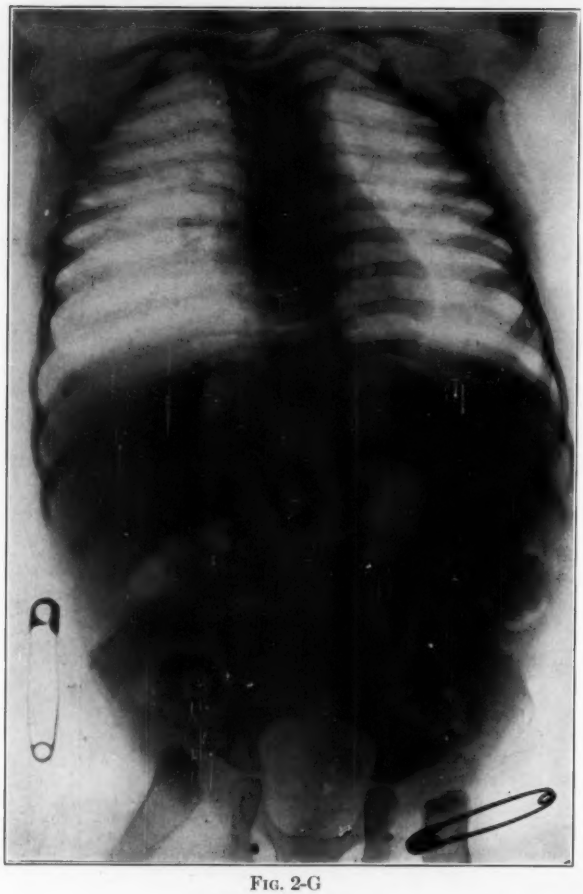

FIG. 2-G
March 3, 1949

In Case 1, the condition was unrecognized until the child was four years of age; roentgenographic examination during the postpartum period revealed the anomaly in Case 2.

Other physical features may make the condition easily recognizable without roentgenograms. The absence of the spinal column below the lumbar region results in obvious narrowing of the pelvis and shortening of the intergluteal fold. Atrophy and dimpling of the buttocks are prominent manifestations, probably due to fibrous-tissue contraction. The muscles of the lower extremities show evidence of atrophy, which becomes increasingly prominent as skeletal growth progresses. Club-foot is a frequent accompaniment.

Photographs of both children reveal the presence of these features. The bony prominence in the lumbar area is easily discernible at the distal end of the spinal column. Narrowing of the pelves, shortening of the intergluteal folds, and dimpling of the buttocks are obvious in both cases. The atrophy of the buttocks, thighs, and calf muscles is more evident in Case 1 (Figs. 1-A and 1-B) than in Case 2 (Figs. 2-A and 2-B).

While little is known concerning the embryological basis of this anomaly, it is interesting to speculate on the etiology.

Ossification of the first sacral vertebra begins at eight to nine weeks of foetal life,

rapidly followed by ossification of the second and third sacral vertebrae. This indicates that the condition must develop during the early days of foetal life. Friedel's theory that minute embryonal trauma results in caudal suppression is the simplest explanation given.

We cannot, however, dismiss the influence of heredity, in view of recent investigations in the field of genetics. Appendicular skeletal anomalies are inherited to a marked degree, and some axial skeletal defects have also been known to have a hereditary basis. No striking genetic discoveries have yet been made in the small number of cases of sacrococcygeal agenesis reported, but there is a good possibility that some hereditary defect is present.

A more remote etiological possibility is that of conjoined twins with dorsal union in the sacral region. One twin may have died in early pregnancy due to poor nutrition, and the other proceeded to term with the persisting defect in the sacral portion of the spinal column.

NOTE: The authors wish to acknowledge the assistance of Robert J. Ayella, M.D., with the photography, and Philip D. Gilbert, M.D., with the x-ray studies.

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TRANSVERSECTOMY FOR SCOLIOSIS

BY H. LESLIE WENGER, M.D., NEW YORK, N. Y.

In 1934, Bisgard was impressed with the almost invariable development of a spinal curvature in patients who had been subjected to a thoracoplasty operation. This spinal curvature occurred with the convexity towards the side operated upon, and the amount of the curvature was correlated with the "relative position of the resected rib to the spine".

The present author found that the development of scoliosis postoperatively was dependent upon the intactness of the transverse processes and the various ligaments attached thereto. Several patients who had had thoracoplasty operations, in which the transverse processes were left undisturbed, strikingly confirmed this conclusion. In other instances, it was noted that pre-existing scoliosis, with the convexity directed toward one side, was corrected when a thoracoplasty, performed for a lung lesion, was done on the opposite side.

On the basis of these observations, a number of patients with primary scoliosis were treated by transversectomies to correct the curvature of the spine.

The following case is reported to demonstrate the efficacy and the simplicity of this method:

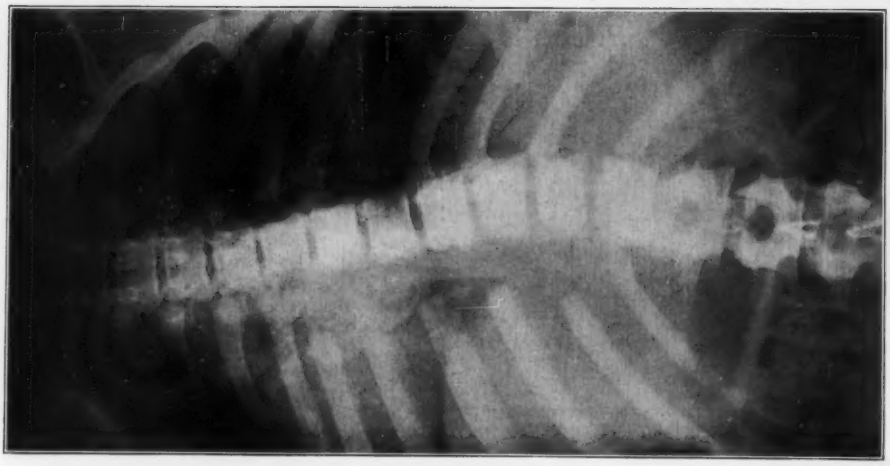

Fig. 3

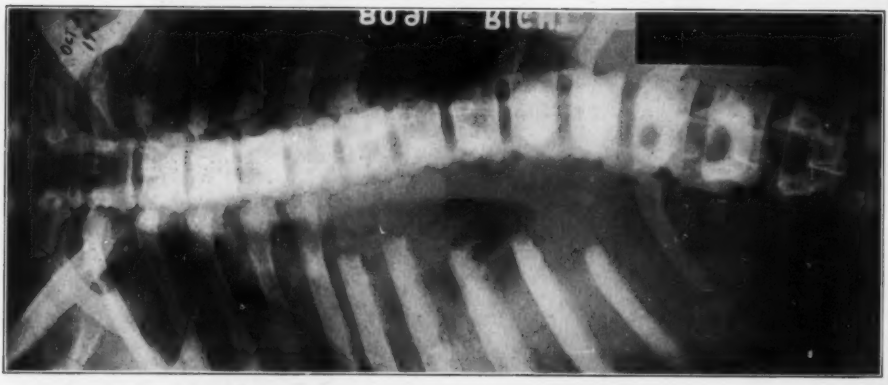

Fig. 2

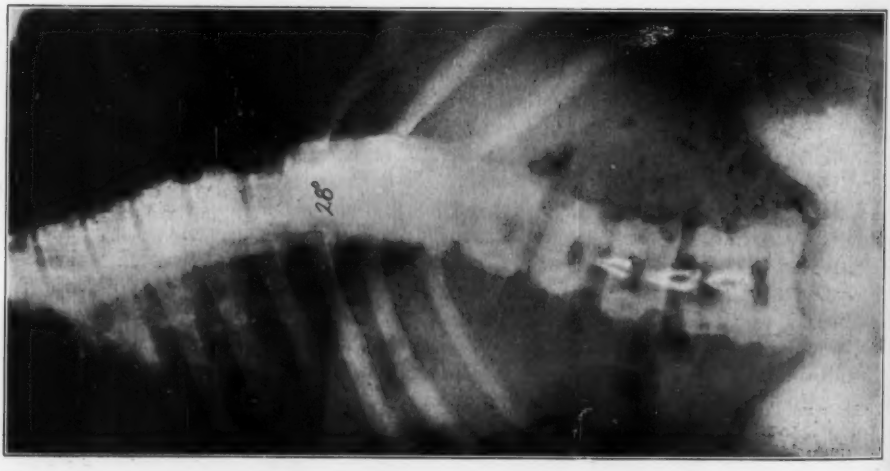

Fig. 1

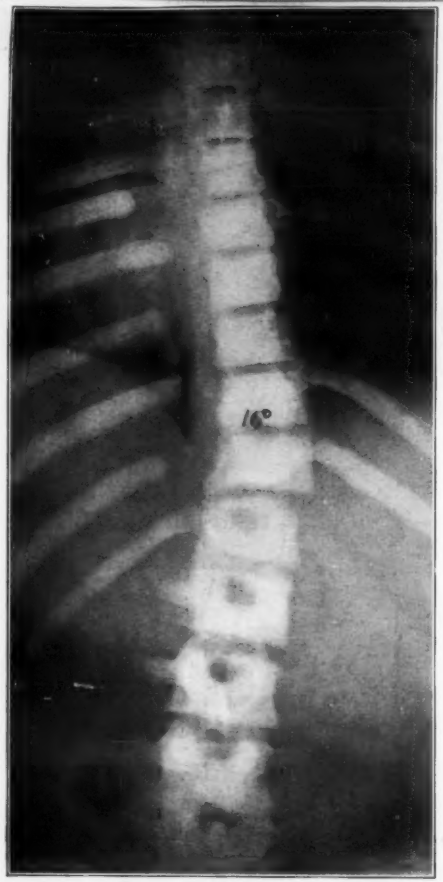

FIG. 4
December 13, 1947

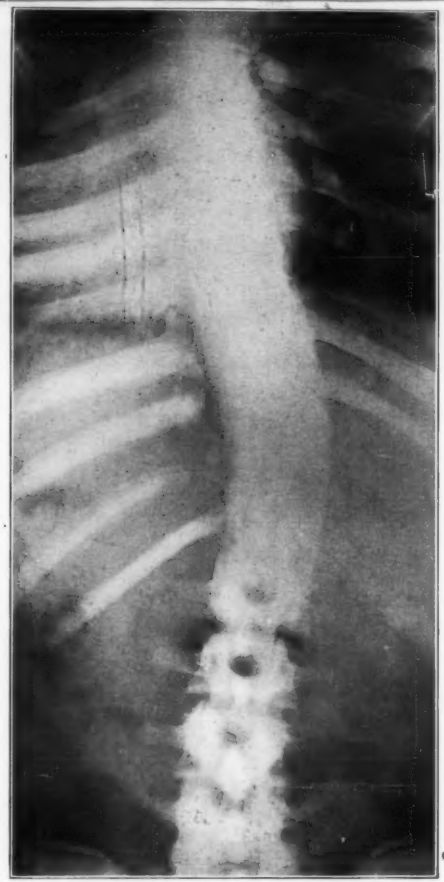

FIG. 5
October 11, 1948

FIG. 6
February 18, 1950

W. S., age sixteen, was first seen on October 13, 1947. Figure 1, a roentgenogram taken at that time, shows a thoracic scoliosis. The curvature was 28 degrees. The operation, performed the next day, consisted of the removal of five transverse processes,—one at the apex of the curve, two from above the apex, and two from below the apex. From two to three inches of the articulating rib was also removed to facilitate the removal of the transverse processes. The patient was discharged following the operation. No plaster-of-Paris correction was used, and the patient was allowed to resume his usual activities. Sixteen days later, on October 29, 1947, a roentgenogram (Fig. 2) was taken which shows the absence of the sections of the articulating rib and the complete extirpation of the transverse processes over the operative site. The roentgenogram shows that there is a correction of the curvature. A further roentgenogram, taken on November 25, 1947 (Fig. 3), shows some regeneration of the ends of the ribs which were operated upon with a maintenance of the correction of the curvature of the spine. On December 13, 1947, two months after the operation, another roentgenogram (Fig. 4) was taken, which shows a maintenance of the immediate postoperative correction; the degree of the curvature then was 16 degrees. (The measurements of the curve were confirmed by Dr. von Lackum.) A roentgenogram taken eleven months later, on October 11, 1948, shows no essential change (Fig. 5); and one taken sixteen months later, on February 18, 1950, shows a maintenance of the improvement (Fig. 6).

SUMMARY

This operation was conceived from observations made on the physiological response of the spine following surgical trauma in thoracoplasties. The above case is reported to demonstrate the efficacy of a very simple operative procedure to assist in the correction of primary thoracic scoliosis. In this instance, a correction of from 28 degrees to 16 degrees was obtained within a period of two weeks following the operation. The correction has persisted to the present day. It was not found necessary to use a plaster cast or any other corrective apparatus.

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LETTER TO THE EDITOR

As Heard At Virginia Beach

(And at other orthopaedic conventions)

How do you pronounce these words so frequently abused by orthopaedic surgeons?

According to the dictionary	As pronounced by orthopaedic surgeons
AB-DO'MEN	AB'DO-MEN
CER'E-BRAL	CER-E'BRAL
DI-A-PHYS'I-AL	DI-APH'Y-SE'AL
EP-I-PHYS'E-AL	E-PIPH'Y-SE'AL
OE-SO-PHAG'E-AL	OE-SOPH'A-GE'AL
HĔM'Ĭ-VERT'E-BRA	HĒ'MĪ-VERT'E-BRA
ME-NIN'GE-AL	MEN'IN-GE'AL
ME-TAS'TA-SIS	MET-A-STA'SIS
OS-TE-O-GEN'E-SIS	OS-TE-O-GEN-E'SIS
OS-TE-OL'Y-SIS	OS-TE-O-LY'SIS
PHA-LAN'GE-AL	PHAL-AN-GE'AL
VER'TE-BRAL	VER-TE'BRAL

Is it worth considering, or don't you care? As you pronounce words, so will your students, house staff, and junior associates.

Anonymous

LIPOMA OF BONE OF INTRA-OSSEOUS ORIGIN *

BY LIEUTENANT ALVIS B. DICKSON, COMMANDER WILLIAM W. AYRES, COMMANDER MALCOLM W. MASON, AND COMMANDER WALTER R. MILLER

Medical Corps, United States Navy

From the United States Naval Hospital, National Naval Medical Center, Bethesda, Maryland

True lipoma of bone of intra-osseous origin, occurring in long bones of the human body, must be quite rare, inasmuch as a search of the literature has revealed only two previously reported cases.

A. S., a thirty-year-old, white, male veteran, was admitted to the United States Naval Hospital on March 3, 1949, with the sole complaint of slight pain in the left ankle, occurring mostly while engaged in mild athletics. This had been present for the past fourteen years, following rather vigorous indulgence in

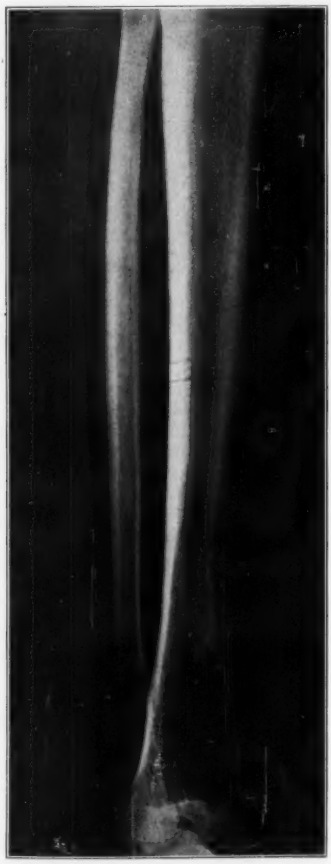

FIG. 1-A

FIG. 1-B

Roentgenograms, March 3, 1949, taken before operation.

* This article has been released for publication by the Division of Publications of the Bureau of Medicine and Surgery of the United States Navy. The opinions and views set forth are those of the authors and not to be considered as reflecting the policies of the Navy Department.

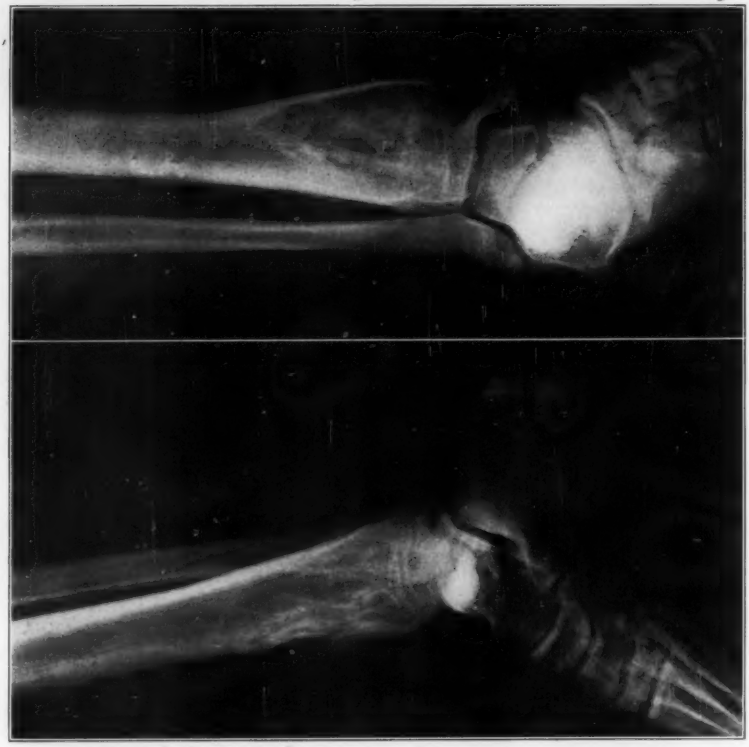

FIG. 3

Roentgenograms, November 30, 1949, taken eight months after operation.

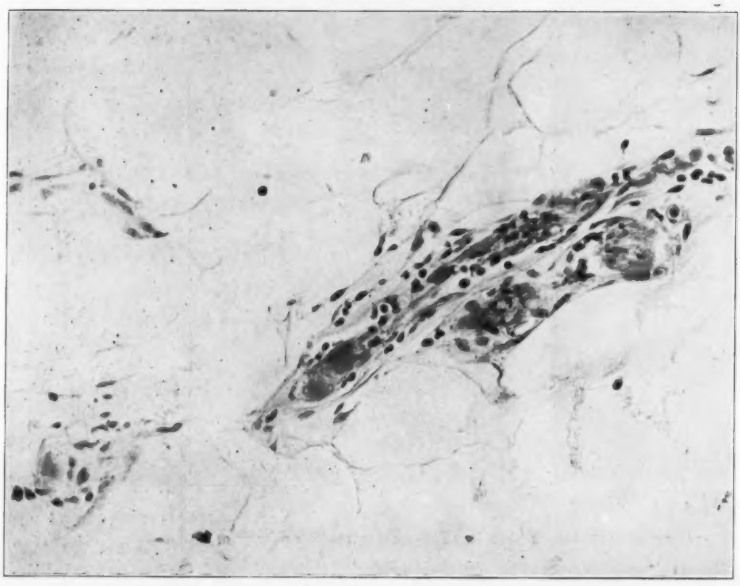

FIG. 2

Photomicrograph (×1000) showing mature fatty tissue in which there were sparse trabeculae of bone.

baseball at the age of sixteen. During the past two years, the pain had become slightly worse and was dull and aching in character; the ankle had become slightly larger than its mate. At no time had there been any evidence of redness or heat, and he had continued at his job, where he walked two to three miles daily.

The physical examination revealed significant findings to be limited to the left ankle which was approximately five-eighths of an inch greater in circumference than the right. There was a complete range of painless motion, no evidence of heat or redness, and only slight discomfort was elicited by firm palpation over the medial aspect of the distal portion of the tibia.

All indicated laboratory procedures, including the Kahn test, gave normal findings.

Roentgenograms of the left leg taken on March 3, 1949, revealed a large oval area of rarefaction, approximately eight by five centimeters in size, situated in the distal end of the tibia, extending from the epiphyseal line upward. There were coarse trabeculations in some portions of the lesion and a slight expansion of the shaft in this area. The general appearance was that of a solitary area of fibrous dysplasia, simple bone cyst, or giant-cell tumor. A complete bone survey of the skeleton revealed no other osseous lesions.

On March 15, 1949, surgical exploration revealed grossly normal periosteum and cortex over the anterior aspect of the lesion. A window cut through the cortex revealed a large cavity, containing yellowish fatty material with gritty particles felt between the fingers. This yellowish material constituted approximately fifteen cubic centimeters in bulk, and glistening oil droplets could be squeezed from it. There were collections of free oil throughout the tumor tissue sufficient to require sponging. The cavity contained no lining, but was surrounded by sclerotic bone. The treatment consisted of thorough curettement and packing with cancellous iliac bone.

For the pathological gross examination, the specimen was obtained by curettage and was found to consist of irregularly shaped fragments of fatty tissue in which there were small fragments of cancellous bone. There was an indication of the breakdown of fatty tissue in that there was much free oil. Also present were a few fragments of normal cortical bone. The total volume of the specimen was approximately fifteen cubic centimeters.

Sections used for the microscopic examination were not decalcified, because of the small amount of bone present. They showed large amounts of mature fatty tissue in which there were sparse trabeculae of bone. The fatty tissue was supported by delicate strands of connective tissue containing capillaries. There was no hemopoiesis. The lipoma was similar to those occurring in subcutaneous tissue, except for the bone trabeculae. When stained with Sudan IV, frozen sections showed fat cells and much extracellular fat in the form of oil globules.

The patient has been examined at intervals of three months and is entirely without symptoms, in spite of spending many of his working hours on his feet. Clinically, he is cured, and roentgenograms reveal progressive filling-in of the defect.

Intra-osseous lipoma occurring in long bones is extremely rare. Rössle mentions only two cases,—one reported by Wehrsig and the other by Brault. Wehrsig described a lipoma of bone in a girl, five and one-half years old. The entire left leg was diffusely thickened. Roentgenograms showed an area of rarefaction of the upper third of the fibula near the epiphyseal line; this third of the fibula was resected. A pathological examination revealed an intra-osseous lipoma, measuring 6 by 5 by 3.75 centimeters, which was divided into three lobules. Brault described an intra-osseous lipoma of the femur.

Although intra-osseous lipoma of the long bones is rare, lipomata occurring in flat bones, particularly of the vertebrae, are not infrequent. When occurring in flat bones, there may be hemangiomatous elements within the fatty tumor. Another type of lipoma involving bone is periosteal lipoma. Geschickter and Copeland report a case of periosteal lipoma of bone in a fifteen-year-old, white female. She gave a history of swelling of one month's duration in the region of the knee joint, and the roentgenogram showed a swelling of the soft parts with slight erosion and roughening of the medial condyle of the femur. Sections showed the typical benign lipoma. Two similar cases have been reported by Bartlett.

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THE USE OF A SPLIT-THICKNESS GRAFT TO COVER THE SKIN DEFECT IN A KRUKENBERG AMPUTATION

BY K. E. KALLIO, M.D., HELSINKI, FINLAND, AND J. E. M. THOMSON, M.D., LINCOLN, NEBRASKA

One of the authors[1], in 1948, published his technique of the Krukenberg operation and reported a series of cases in which this method had been employed during and after World War II. This lobster-claw type of amputation has proved particularly useful in the rehabilitation of persons who have lost both hands or the greater parts of both hands, because the holding surfaces of the two branches have touch sensation and give assurance for many functional activities.

To maintain sensation, the skin incision is so devised that the contact surfaces of the radial and ulnar branches are covered with the patient's own skin. This leaves a sizable defect, the full length of the dorsal aspect of the ulnar branch. In the past, this defect was covered by a pedicle skin graft from the abdomen, so that weeks of immobilization were required before the pedicle could be freed; the resulting atrophy from disuse of the muscles attached to the ulnar and radial branches materially delayed rehabilitation and influenced the active function and grasping power in the stump.

In August 1948, the suggestion was offered by Thomson that, with a dermatome, a split-thickness skin graft could be applied to this ulnar defect, allowing function of the branches to be started a few days after operation,—even before the pressure dressing was removed.

A favorable case for a Krukenberg operation was that of J. K., a worker, sixteen years of age. Both hands had been severely damaged in an industrial accident, several months earlier. The left hand and distal portion of the forearm had been amputated, leaving a healed stump 19 centimeters in length. On the right hand there remained the thumb, the second metacarpal, and stumps of the third and fifth metacarpals. A previous plastic procedure had given him a functional grasping surface between the first metacarpal and the thumb.

On August 12, 1948, a Krukenberg operation was performed on the left stump. A split-thickness skin graft was procured with a dermatome from the dorsum of the right thigh to cover the ulnar defect. On the fifth day, dressings were changed and active motion of the branches was started. These efforts were, of course, minimal at first, because protective dressings were required over the wounds. After the stitches had been removed at the end of ten days, a program of active rehabilitation was started; the patient left the hospital on September 19. At that time the opening angle of the branches was 20 degrees and the speed was seventy-six times a minute (Figs. 1-A and 1-B).

FIG. 1-A

FIG. 1-B

J. K. Appearance of pinchers and split-thickness skin graft on ulnar branch September 8, 1948, less than one month after operation.

Fig. 1-A: Note the extent of loss of right hand as the thumb presses against the radial branch of the left forearm.

Fig. 1-B: Showing the range of active spreading of the branches.

FIG. 1-C

FIG. 1-D

Shows active spreading and closing of the branches of Krukenberg stump in April 1950, twenty months after surgery. The function of the right hand is also shown.

The patient has not been seen by the authors since he left the hospital, but he reported on April 4, 1950, that he uses his Krukenberg stump for all purposes, that he has direct, hinge-like movement to an opening angle of 30 degrees with no discomfort; that the split-thickness skin graft has some sensation, although it is not so acute as the skin on the other parts of the branches; and that the skin now looks almost the same as normal skin.

Since the experience with this case, a dermatome split-thickness skin graft has been used in every Krukenberg operation with excellent results and no failures of the graft to take.

A split-thickness skin graft to cover the dorsal ulnar skin defect of the Krukenberg operation has the following advantages:

1. It simplifies the operative technique by eliminating the need of an abdominal pedicle graft.

2. It is much more comfortable for the patient to have his arm free during healing of the wound.

3. It enables early use and rehabilitation of the branches, thus providing an economic advantage.

4. The graft assumes a very normal skin appearance and develops a measure of sensitivity.

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OSTEOCHONDRAL FRACTURE OF THE LATERAL FEMORAL CONDYLE

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A tangential osteochondral fracture of the patella is an infrequently reported condition. It is generally agreed that the mechanism of injury is due to a lateral dislocation of the patella, which hooks itself over the lateral femoral condyle. Upon contraction of the quadriceps, the patella is pulled back into place; if the patella does not clear the projecting femoral condyle, a portion of its articular surface is sheared off, producing the typical osteochondral patellar fracture.

In all cases previously reported where the forces involved have been indirect and purely muscular, the patella has borne the brunt of the injury. The trauma to the lateral femoral condyle has been slight and incidental. In the case here presented, it is clear that the inferolateral border of the lateral femoral condyle was knocked out by the spontaneous reduction of the dislocated patella, and an osteochondral fracture of the femoral condyle was produced.

CASE REPORT

A. R., a well-developed student, aged twenty-four years, was in the habit of exercising with a double-spring "muscle developer". One evening he fitted one handle of the "developer" onto his left foot, holding it with his right hand, and holding the other grip in his left hand, stretched upward and outward with the springs under tension. He stumbled, causing the left leg, which was already extended, to be forcibly abducted and externally rotated. Sudden severe pain was felt, and the knee became locked in extension. The patella was visibly dislocated and could be felt on the lateral side of the knee joint, and the tibia appeared externally rotated on the femur. The patient attempted reduction by grasping his leg with one hand and his thigh with the other, rotating the tibia inward on the femur. The patella sprang back into position with a loud crack, and active knee movement was resumed.

Following the incident, the patient complained of frequent "locking" and "giving way" of the left knee. A marked swelling appeared. He first reported to this Hospital seven days after the injury. There was no previous history of disability or of injury to the knee.

Palpation revealed that the knee joint was filled with fluid. Ninety cubic centimeters of blood was aspirated from the joint. There was a marked tenderness over the lateral femoral condyle and over the medial border of the patella. A mobile loose body was felt on the medial aspect of the patella. There was no ligamentous instability; neither was there any genu valgum deformity.

A roentgenogram showed a crescent-shaped, loose body in the knee joint on the inferomedial aspect of the patella (Figs. 1-A and 1-B) which was consistent with a triangular bony defect on the lateral aspect of the lateral femoral condyle (Fig. 1-C). There was no doubt that the loose fragment originated from the femoral condyle.

Three weeks after the original injury, under spinal anaesthesia, the knee joint was opened by a medial parapatellar incision eight centimeters in length. A large amount of blood-stained fluid and many clots were removed from the joint. A loose, crescent-shaped body immediately presented itself and was lifted out. Its convex surface was covered with smooth, glistening cartilage, while its concave surface was covered with a thin layer of cancellous bone. Apart from some roughening of the medial articular surface of the patella, there were no defects in the knee joint. It was not possible to redislocate the patella, but, as a prophylactic measure, the capsule was plicated during closure of the joint.

Convalescence was uneventful, and the patient was discharged from the Hospital ten days later. Within one month after the operation, he had full knee movement and no disability apart from a slight aching around the joint.

The pathological report was as follows: "The specimen is a fragment of bone and cartilage of somewhat elliptical shape and measuring 3 by 2 by 0.5 centimeters. The convex surface presents a smooth white cartilage and the concave surface is rough and bony. Microscopic examination reveals a necrotic bony base on one side and a viable cartilage on the other side. The cartilage at one end is fissured and shredded into thick fibers which merge into connective tissue. The connective tissue shows infiltration of numerous lymphocytes and many congested blood vessels. The marrow between the trabeculae of the necrotic bony base contains organized connective tissue, lymphocytes, histiocytes, a few granulocytes, and numerous fibroblasts.

"A section taken from the center of the fragment shows similar changes, but here the subchrondral bone is viable and there are hemorrhagic areas scattered within the marrow. Diagnosis: Osteochondral fracture." *H. Ungar, M.D.*

DISCUSSION

The precise and definite nature of this case history establishes the fact that a forced abduction-external rotation injury at the knee joint caused a relatively stable patella to dislocate outward. In the circumstances described, the quadriceps and particularly the

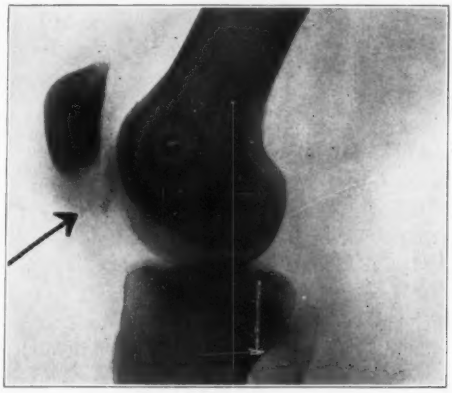

FIG. 1-A

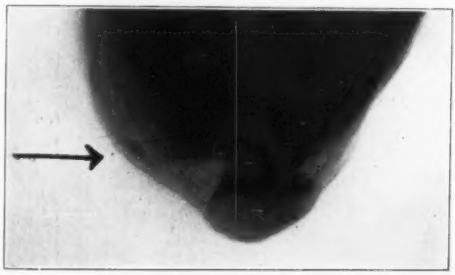

FIG. 1-B

FIG. 1-C

Fig. 1-A: Lateral view of the knee joint. Osteochondral fracture of lateral femoral condyle. Roentgenogram shows a crescent-shaped bone fragment loose in the joint.

Fig. 1-B: "Skyline view". Osteochondral fragment loose in the medial parapatellar region of the knee joint.

Fig. 1-C: Anteroposterior view of the knee joint. The bony defect of the inferior aspect of the lateral femoral condyle is caused by the shearing off of the fragment.

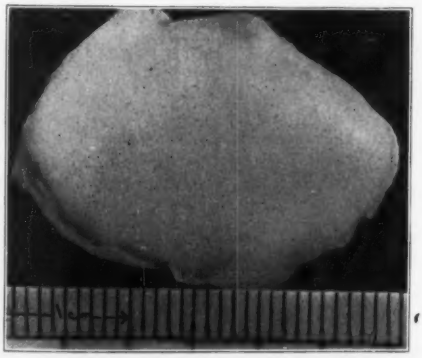

FIG. 2-A

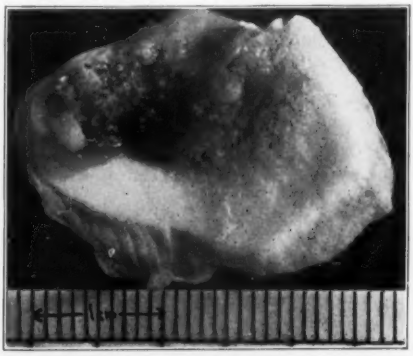

FIG. 2-B

Fig. 2-A: Convex surface of the osteochondral fragment is smooth and cartilaginous.
Fig. 2-B: Fractured surface of the osteochondral fragment is covered by a thin layer of cancellous bone.

vastus internus were subjected to tension and spasm. The patient reversed the process of injury by internally rotating the tibia on the femur. This gave enough slack for the vastus internus to pull back the patella across the lateral femoral condyle. If the patellar fails to clear the projecting femoral condyle, damage will occur either to the patella itself or to the femoral condyle. If damage occurs to the patella, an osteochondral patellar fracture results; while, if damage occurs to the femoral condyle, the type of injury is as here described. The mechanism of injury is identical in both cases.

The pathological report clearly confirms that the loose body was traumatic in origin and not to be confused with a loose body resulting from osteochondritis dissecans.

Harmon reported a traumatic osteochondral fracture of the lateral femoral condyle which was complicated by an open wound of the knee joint. This fracture was the result of direct external trauma. However, in the case of the fracture herein described, the causative factors were indirect and purely muscular.

It is of interest here to note that McFarland, in his report of six consecutive cases of recurrent dislocation of the patella, described the fragmentation and damage both to the lateral femoral condyle and to the patella. It would appear, therefore, that even when the lateral femoral condyle tends to be underdeveloped and the traumatic factor is minimal, the condyle suffers damage by the shearing action of the sliding patella. It follows then, that, with a well developed lateral femoral condyle and a stable patella, dislocation, being more difficult to produce, is correspondingly more difficult to reduce, and greater damage (such as an osteochondral flake fracture) may be expected.

Milgram reviewed the literature of osteochondral patellar fracture, defined the mechanics of injury, and listed the condition as a definite clinical entity. He reported six operative cases. In three of these, in addition to the patellar fracture, there was clear evidence of injury to the lateral femoral condyle; two cases presented shearing off of the superficial layers of cartilage; and the third showed the fissured lines of a stellate fracture of the condyle.

Coleman reported two cases of recurrent osteochondral patellar fracture, in each of which there was a scraping and a scoring of the articular cartilage of the lateral femoral condyle. Coleman confirmed Milgram's conception of the mechanics of injury.

Kroner, in 1904, was the first to report osteochondral flake fracture in the knee joint. In his case, and in all cases subsequently reported, the major injury was sustained by the patella. The present case presents the unusual feature of an osteochondral fragment originating from the lateral femoral condyle.

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NOTE ON THE USE OF NITROGEN MUSTARD IN RHEUMATOID ARTHRITIS

With Consideration of the Similarity of Its Effect to That of Cortisone and ACTH

BY NED M. SHUTKIN, M.D., DURHAM, NORTH CAROLINA

From the Orthopaedic Division, Department of Surgery, Duke University School of Medicine, Durham

In 1946, Gilman and Phillips reported on the biological actions and the possible therapeutic applications of mustard-gas compounds. Research on chemical-warfare agents had demonstrated the systemic cytotoxic action of mustard gas and its related compounds, in addition to its property as a topical vesicant. The systemic cytotoxic action is especially characteristic of the nitrogen and sulphur derivatives; it chiefly affects proliferative tissues, particularly those of a lymphoid nature. In addition, a lymphopaenic effect has been demonstrated.

The fact that rheumatoid arthritis entails an active proliferation of the synovial membrane, with perivascular lymphocytic infiltration, and perivascular lymphocytic nodules in the subcutaneous tissues, endomysium and perimysium, and perineural tissues [2, 4, 11], prompted investigation of the effect of nitrogen mustard in this disease.

Six cases of active rheumatoid arthritis were studied.* The prescribed dosage of 0.1 milligram of methyl-bis (Beta chloro-ethyl) amine hydrochloride per kilogram of body weight was administered intravenously on four consecutive days. The responses to the drug were transient, but, in all instances, they were great enough and occurred promptly enough to suggest a possible cause-and-effect relationship.

Of interest in connection with these findings are recent studies of the adrenal cortex steroid hormones and the adrenocorticotropic hormone of the pituitary (ACTH) [1, 5, 6, 7]. The most abundant sterol in vertebrates is cholesterol, which is synthesized in the body and which, in all probability, is the source of the steroid hormones. There is abundant cholesterol in the adrenal cortex, about 90 per cent. of which is esterified with fatty acids. To date, some twenty-six steroid hormones have been isolated from the adrenal cortex, but only three groups appear to have biological function. Compounds E and F of Kendall are in one of these groups. The greater part of these compounds are degraded in the body into carbon dioxide and water, and only small amounts are excreted in the urine. These steroids are secreted as the result of the stimulation of the adrenal cortex by ACTH. Administration of ACTH rapidly depletes the adrenal cortex of its esterified cholesterol, which presumably is converted into steroid hormone. That this constitutes a specific response is indicated by the fact that no other organ evinces a fall in cholesterol content upon ACTH administration. Furthermore, an assay of blood obtained from the renal vein in the experimental animal after administration of ACTH shows a corresponding rise in the hormones.

Another unique feature of the adrenal cortex is its high content of ascorbic acid. Interestingly enough, this compound also shows rapid depletion after the administration of ACTH. Thus, two specific effects of ACTH upon the adrenal cortex are the depletion of cholesterol and of ascorbic acid.[10]

The various physiological concerns of the adrenal-cortical hormones have been listed [7] as follows: (1) resistance to stress; (2) regulation of electrolyte and water balance; (3) regulation of carbohydrate and protein metabolism; (4) effect upon work capacity of skeletal muscle; (5) alteration in permeability; and (6) control over the lymphocyte,

*This work was done under a fellowship of The National Foundation for Infantile Paralysis, Inc.

monocyte, and eosinophil level in the blood. Only one of these concerns us here: the leucopaenic effect of the hormones, an effect also produced by the administration of nitrogen mustard.

It is of interest to note that the findings of Ludewig and Chanutin indicate that the administration of nitrogen mustard to rats produces an appreciable decrease in the adrenal ester cholesterol and a mild decrease in the ascorbic acid within the first twelve hours. It should be noted that lethal doses of nitrogen mustard were given. However, the response of depletion of cholesterol and ascorbic acid was prompt, whereas the toxic manifestations did not occur until the fourth day.

These findings suggest that the beneficial effects following the administration of nitrogen mustard may be due to action either directly upon the adrenal cortex or upon the pituitary gland with a subsequent secretion of ACTH.

A further parallel is found in the fact that nitrogen mustard has a lymphopaenic effect, as do ACTH and cortisone.

Nitrogen mustard is not recommended as a therapeutic agent in rheumatoid arthritis. The unpredictability of the disease with its tendency to abrupt but temporary remissions, the possible beneficial effect of the rest and respite from responsibility occasioned by mere hospitalization, the misguided optimism of the arthritic who has already run the therapeutic gamut, and, perhaps, the unbridled enthusiasm of the investigator, all tend to compromise the validity of the findings. Nor can it be determined whether the objective evidence of improvement, such as increased range of motion, decreased periarticular-skin temperature, or decreased effusion, is related to a course of medication or merely coincident with it. The difficulty of differentiating mere coincidence from a cause-and-effect relationship has been attested in recent years by the short-lived popularity of colloidal sulphur, bee and cobra venoms, vitamin D, colloidal gold, and other drugs. These compounds, on continued clinical use, failed to maintain the therapeutic standards for which they were heralded in the initial reports. However, the similarity in biochemical and clinical response to ACTH and nitrogen mustard justify further investigation of the latter agent in the therapy of rheumatoid arthritis.

SUMMARY

1. Six patients with active rheumatoid arthritis showed prompt, but transient, improvement after the administration of nitrogen mustard.

2. Nitrogen mustard produces depletion of the cholesterol esters and ascorbic acid of the adrenal cortex in the experimental animal.

3. ACTH produces depletion of the cholesterol esters and ascorbic acid of the adrenal cortex.

4. Further investigation to determine the efficacy of nitrogen mustard in the treatment of rheumatoid arthritis is indicated.

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AN IMPROVED ABDUCTION EXERCISE SPLINT FOR THE SHOULDER

BY EDWARD J. COUGHLIN, JR., M.D., WILLIAMSTOWN, MASSACHUSETTS

In April 1949 the author published an article[1] entitled "An Abduction Exercise Splint for the Shoulder". The author did not claim credit for devising the splint, but mentioned that the original model had been seen by him at a prisoner-of-war camp in southern France. As the result of the European circulation of *The Journal*, the author has been in correspondence with the originator of the apparatus, Dr. Georg Neubauer, of Graz, Austria. Dr. Neubauer has been kind enough to forward his own modifications of the original splint, which present some improvements over that described by the author.

In the modification of the original splint (Figs. 1 and 2), the thorax is supported on

Fig. 1

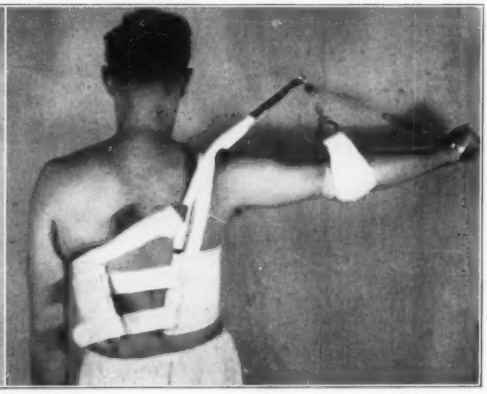

Fig. 2

both sides, which gives more adequate fixation to the apparatus and prevents it from moving in the process of abduction. In this way, one gets the maximum degree of abduction at the shoulder without untoward motion of the apparatus itself.

The author takes this opportunity to thank Dr. Neubauer, not only for his original idea of the splint, but also for his kindness in sending illustrations of the more recent modification.

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AN IMPROVED SPINE RETRACTOR

BY CHARLES H. FRANTZ, M.D., GRAND RAPIDS, MICHIGAN

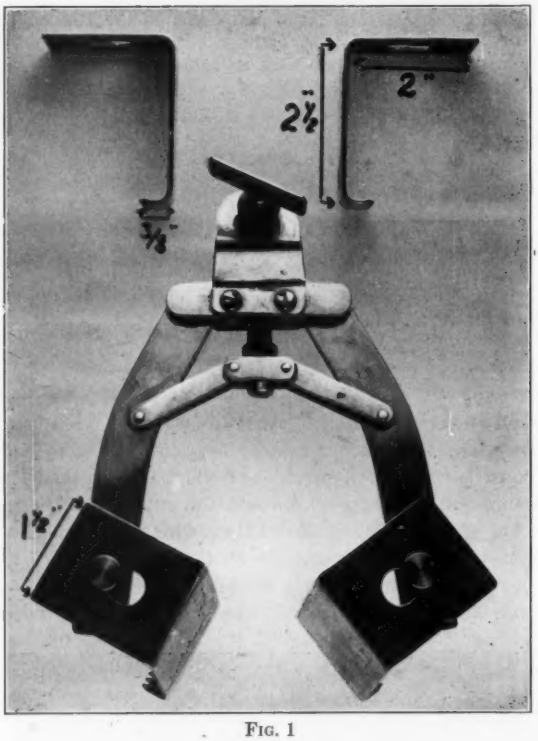

FIG. 1

Jensen,[1] in 1949, fabricated a spine retractor from a medium-sized Kirschner clevis, utilizing the principle of Hibbs in the blades. This retractor is very strong and is easily constructed. However, the turning handle is difficult to manipulate after the retractor is in place, and necessitates raising the clevis out of the operative field. The blades do not curve sufficiently to grasp the lateral muscle masses, and tend to rise out of the operative field.

The following modifications have proved to be satisfactory:

1. The turning handle has been removed and a beveled (45-degree angle) stainless-steel gear has been placed at right angles to the turning screw (Fig. 2). The mechanical advantage is thus increased twofold. A stainless-steel housing covers the mechanism.

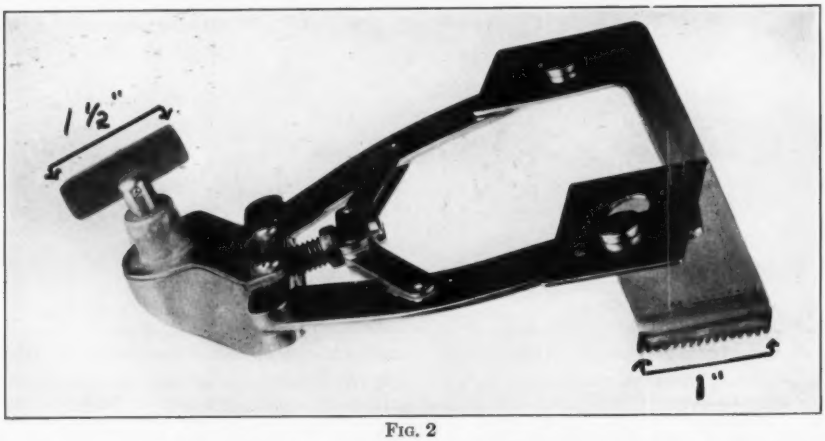

FIG. 2

2. The curve of the blades has been increased to three-eighths of an inch, which tends to fix the blades more securely under the muscle masses.

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BRACE TO CORRECT VARUS OF THE FORE PART OF THE FOOT

BY HAROLD LUSSKIN, M.D., NEW YORK, N. Y.

In the last few years several cases of varus of the fore part of the foot have come to the author's attention, some of them as primary conditions, others as residual deformities of club-foot. In several of these cases plaster casts were required when the patients were over a year old; in others the deformity could be controlled by a brace.

The brace which has been used is made of leather and stainless steel (Figs. 1-A and 1-B), and is attached to the patient's foot by means of two straps (Fig. 2). By adequate bending of the long arm of the brace, it is possible to increase the correction until the foot is in valgus.

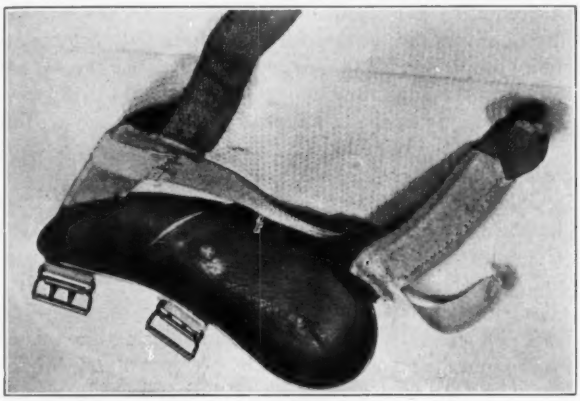

FIG. 1-A

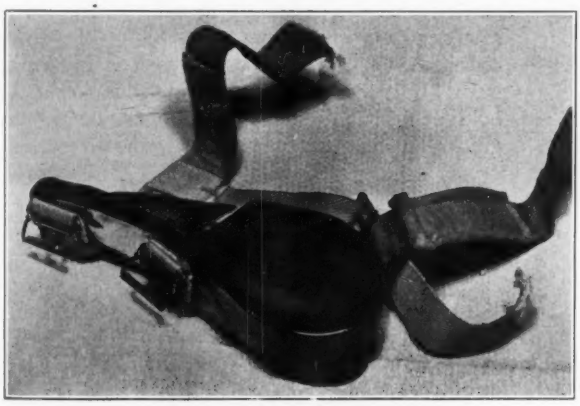

FIG. 1-B

FIG. 2

Figs. 1-A and 1-B: Photographs of the brace as seen from above (Fig. 1-A) and from below (Fig. 1-B). This brace consists of a J-shaped piece of stainless steel, placed with the long arm on the lateral aspect of the foot and the short arm passing around the heel. A small extension from the long arm passes underneath the foot. A leather mold fills this metal reinforcement, into which the foot fits. Two straps are fastened to the brace, one passing around the ankle and the other around the fore part of the foot.

Fig. 2: The brace is attached to the foot. It is advisable to use a thin felt between each strap and the skin. Where the front strap tends to slip off the big toe, as in short feet, the front and rear straps are connected by another band on the inner side of the foot.

Primarily, this brace is to be used in conjunction with strapping, or it may serve as a night brace when strapping or other forms of therapy have been discontinued. It can be used whenever the child is not standing.

CIRCULAR HAND SAW FOR CUTTING SMALL BONES

BY EMANUEL B. KAPLAN, M.D., NEW YORK, N. Y.

When, in the course of an operative procedure, the necessity arises to cut through small bones, the osteotome is doubtless the instrument of choice. In certain circumstances a wire saw or a hand saw can be used, but such an instrument is not always practical in narrow operative fields, as, for instance, between the metacarpals or metatarsals. It was thought that, for this and other reasons, a circular hand saw with some protection for the surrounding tissues could be used with great advantage. It was further considered that the ring cutter used by the jewellers could be properly adjusted for surgical purposes.

After several unsuccessful attempts, an instrument was constructed which proved useful in transverse osteotomies of the metacarpals, metatarsals, phalanges, the proximal end of the radius, distal end of the ulna, the clavicle, the fibula, the ribs, and even the transverse processes of the thoracic and lumbar vertebrae.

The instrument is made of stainless steel; its length is about nine inches. It consists of a long rod with an appropriate handle at one end; a circular saw is placed in a slot at the opposite end of the instrument (Figs. 1 and 2). The circular saw is made of very resistant steel and is about one and one-quarter inches in diameter. The plane of the saw is vertical and coincides with the long axis of the instrument. About two inches back from the center of the saw, a metal piece is affixed on a transverse axis, opening and closing like scissors. This piece has an arrangement for the thumb and can be closed by pressure of the thumb of the hand which also holds the instrument. As soon as pressure has been released, this piece opens, because of a spring. The attachment is extended forward and downward to form a protective jaw under the circular saw. Releasing the thumb opens a space between the saw and protective jaw; pressure by the thumb closes the space. The protective jaw is semicircular, to fit the curve of the saw. It is serrated to hold the bone and has a shallow longitudinal groove. When closed, the saw fits the longitudinal groove of the jaw.

FIG. 1

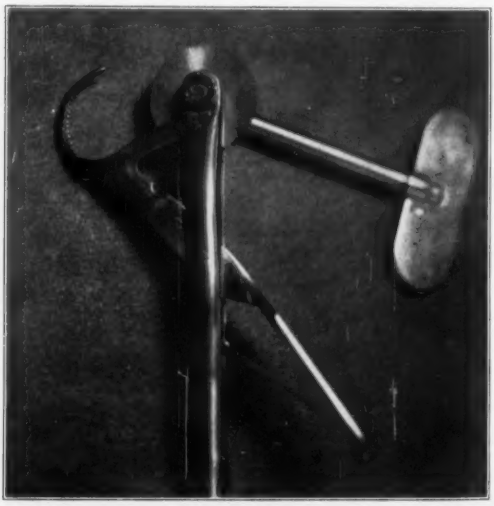

FIG. 2

The serrations alone have proved insufficient to prevent rotation of the bone and, therefore, the instrument has a metal tongue which engages into the slot behind the center of the saw; when the instrument is closed, this tongue steadies the bone for the osteotomy. The circular motion of the blade is produced by a handle, three and one-half inches long, attached to the axis of the blade on the right side of the instrument. This handle manipulates on a universal joint and can be worked from several angles. A holder for the fingers makes it easier to rotate.

After the location of the osteotomy has been exposed, the instrument is held in one hand and the protective jaw is carefully engaged under the bone. Pressure is gently applied to the thumb piece until the saw touches the bone over the site of the osteotomy. Firm, controlled pressure is maintained by the thumb of the hand which holds the instrument; with the other hand the surgeon rotates the handle connected with the blade until the bone is sawed through.

The instrument has the following advantages:

1. Hammering is eliminated.
2. The circular saw produces an even, neat line of division.
3. The protective jaw prevents injury to the soft tissues.
4. It can be placed safely in comparatively inaccessible locations, where a wire saw or the usual hand saw would not fit.
5. It is easy to manipulate and can be sterilized and cleaned easily.
6. It can be used in either hand.

ANNOUNCEMENT

As Corresponding Editors, a number of distinguished orthopaedic surgeons of many countries have recently accepted appointment on the staff of *The Journal*. They will serve as representatives of the orthopaedic surgery of their countries for the British and American Volumes. Through their reports our readers may be informed of the proceedings of the meetings of their societies, of honors which have been conferred, and of the elections of presidents and other officers. One may keep abreast of educational developments in the orthopaedics of other countries, and, finally, of scientific and clinical progress through résumés of important papers presented at their meetings.

In 1927, Jacques Calvé of Berck-Plage and Murk Jansen of Leiden became Foreign Editors of *The Journal*. During the same year, there were added other illustrious surgeons,—Patrik Haglund of Stockholm, Way-Sung New of Shanghai, Hans Spitzy of Vienna, Fritz Lange of Munich, and Y. Tashiro of Tokyo. In 1928, Vittorio Putti joined the editorial staff as Foreign Editor for Italy, and in 1930, Bedrich Frejka, of Brno, for Czechoslovakia. Many, interesting, and of inestimable value were the contributions of these men. With the suspension of the mails in 1940, and the intense preoccupations which followed, the publication of proceedings of orthopaedic societies in other countries was no longer possible.

We are glad to announce that the following will serve as Corresponding Editors of *The Journal*:

Merle d'Aubigne, France
José Luis Bado, Uruguay
Walter Ehalt, Austria
Juan Farill, Mexico
Sten Friberg, Sweden
F. E. Godoy Moreira, Brazil
Adam Gruca, Poland
Alberto Inclan, Cuba
K. E. Kallio, Finland
Johannes Mortens, Denmark
J. D. Mulder, Netherlands
Arnaldo Rodo, Portugal
José Valls, Argentina
Jean Verbrugge, Belgium

Through their cooperation, we may confidently look forward to a broadening of our contacts with the orthopaedic surgery in other countries.

William A. Rogers

News Notes

The Eighteenth Annual Meeting of **The American Academy of Orthopaedic Surgeons** will be held at the Palmer House, Chicago, Illinois, from January 27 to February 1, 1951, under the presidency of Dr. Guy A. Caldwell.

The Annual Meeting of **The American Orthopaedic Association** will be held at The Greenbrier, White Sulphur Springs, West Virginia, on June 20, 21, 22, and 23, under the presidency of Dr. James S. Speed.

The Newington Home and Hospital for Crippled Children, Newington, Connecticut, has announced an approved one-year resident-fellowship in children's work, starting July 1, 1951.

The Fifth Congress of the **International Society of Orthopaedic and Traumatic Surgery** will be held in Stockholm, Sweden, May 21 to 25, 1951. The Secretary of the Congress is Dr. Anders Karlen, Karolinska Institutets Ortopediska Klinic, Stockholm.

The two main subjects will be: (1) Avitaminosis and Bone Changes, with Dr. R. Zanoli of Italy and Dr. E. Rutishauser of Switzerland as the principal speakers; and (2) Low-Back Pain and Sciatica. Results of Treatment, with Dr. Joseph S. Barr, of Boston, Massachusetts, as the first speaker. In addition a large number of miscellaneous papers will be presented.

The Fifth Annual **Amputee Conference** will be held at the **Kessler Institute for Rehabilitation,** West Orange, New Jersey, April 5 to 7, 1951. The Conference is open to doctors, limb makers, physical therapists, and other rehabilitation personnel.

The program will consist of lectures and demonstrations by the staff and visiting specialists and will be devoted not only to discussions of the anatomy and basic surgery of the extremities, but also to demonstrations of prostheses, and discussions of the training and rehabilitation of the amputee.

The Fifth Congress of the **Pan-Pacific Surgical Association** will be held in Honolulu from November 10 to 21, 1951, under the presidency of Dr. F. J. Pinkerton.

The object of the Pan-Pacific Surgical Association is to bring together surgeons from countries bordering on the Pacific Ocean in order to permit the exchange of ideas and methods and to develop a spirit of good fellowship among the various races represented. Although it was originally planned to hold meetings every three years, world events have permitted but four conferences since the establishment of the organization,—the first in 1929 and the last in 1948.

Further information may be obtained by writing to the Pan-Pacific Surgical Association office, Suite 7, Young Building, Honolulu, Territory of Hawaii.

The Fifth Annual Meeting of the **New Jersey Orthopaedic Society** was held at St. Francis Hospital in Trenton, New Jersey, on October 12, 1950. The following program was presented:

The Conservative Treatment of Trochanteric Fractures: A. M. Rechtman, M.D.
Complications in Surgery of Hip Fractures: O. R. Carlander, M.D.
Arthropathies: W. R. Peterson, M.D.
The Combined Operation in Disc Surgery: W. Scheuerman, M.D. (By invitation.)
The Problems of Pathological Fractures: O. Lehman, M.D.
Interesting Bone Tumors: A. S. Thurm, M.D.
Officers for the ensuing year are:

President, Nicholas S. Ransohoff, M.D.
President-Elect, Raphael R. Goldenberg, M.D.
Secretary, Philip Willner, M.D.
Treasurer, Roy R. Ciccone, M.D.

The **November** issue of the **British Volume** of *The Journal of Bone and Joint Surgery* contains special articles by American, Canadian, and British surgeons, reviewing the progress of orthopaedic surgery for the last fifty years. Due to various delays, the November issue may not reach *Journal* readers until after the January 1951 issue. The latest word received from the London office is to the effect that the November number will be distributed to British subscribers on January 20. If this is possible, it should reach American subscribers by February 15 or 20.

The Second European **Congress of Rheumatology** of *La Ligue Europeenne contre le Rhumatisme*, planned by the host society, *La Sociedad Española de Reumatismo*, will be held in Barcelona, Spain, September 24 to 27, 1951. All doctors interested in the treatment of rheumatism are invited to attend.

Among the subjects to be discussed at the Congress are:

Non-articular rheumatism, the effects of pituitary and adrenal-cortex hormones in the treatment of rheumatic diseases, suprasonic therapy, ankylosing spondylitis, and brucellar rheumatism.

La Sociedad Española de Reumatismo invites all doctors who are interested in these subjects to submit papers for discussion at the meetings of the Congress. Information regarding the length of papers and the conditions under which they may be submitted may be obtained from the Planning Committee of the Congress (*Comité organizador del Congreso*).

In addition, *La Sociedad Española de Reumatismo* plans as a feature of the Congress a bibliographical exhibition of all works on rheumatism written during the last twenty years. The Society invites authors who are interested in being represented in such an exhibit to send a copy of each of their books or monographs to the Planning Committee of the Congress.

Requests for further information about the Congress should be sent to Dr. Pedro Barcelo, General Secretary of the Congress, c. Casanovas, 209, Barcelona, Spain.

The **National Foundation for Infantile Paralysis** announces the availability of a number of predoctoral fellowships, covering a period of one year, and postdoctoral fellowships, covering a period of from one to three years, to candidates whose interests are in research and teaching in the fields related to the problems of poliomyelitis,—such as virology, biochemistry, biophysics, orthopaedics, pediatrics, neurology, and epidemiology.

Complete information regarding these fellowships and applications may be obtained from the Division of Professional Education, National Foundation for Infantile Paralysis, 120 Broadway, New York 5, N. Y.

The **American Society for Surgery of the Hand** will hold its Annual Meeting in Chicago, January 26 and 27. The sessions on Friday will be held at the Palmer House. Registration will begin at 8:30 and the following program has been planned:

Morning Session (9:00 o'clock)

Internal Splint for Closed and Open Treatment of Injuries of the Extensor Tendon at the Distal Joint of the Finger. Donald R. Pratt, M.D., San Francisco, California.

Repair of the Extensor-Tendon Insertions in the Fingers. H. Minor Nichols, M.D., Portland, Oregon (by invitation).

Cerebral Palsy As It Affects the Upper Extremities. Kenneth B. Jacques, M.D., Los Angeles, California (by invitation).

Surgery of the Upper Extremity in Cerebral Palsy. William Cooper, M.D., New York, N. Y. (by invitation).

Arthritic Deformities of the Hand: Their Prevention and Treatment. Arthur Steindler, M.D., Iowa City, Iowa.

Afternoon Session (1:30 o'clock)

Arthrodesis in the Hand. William H. Frackelton, M.D., Milwaukee, Wisconsin.

Electromyograms. James G. Golseth, M.D., Pasadena, California (by invitation).

Reconstructive Surgery on the Upper Extremity in Poliomyelitis. C. E. Irwin, M.D., and Don Eyler, M.D., Warm Springs, Georgia (by invitation).

The Correction of Some Congenital Hand Deformities. Jerome P. Webster, M.D., New York, N. Y.

Methods of Treatment of Neuromata of the Hand. Gordon H. Grant, M.D., Victoria, British Columbia (by invitation).

At 7:00 o'clock, dinner and a business meeting for members will be held at the University Club. The program on Saturday morning, for members of the Society, will be held at the Passavant Hospital.

PAUL PLUMMER SWETT
1882–1950

Dr. Paul Plummer Swett of Bloomfield, Connecticut, was born in Granville, Massachusetts, August 23, 1882, and died at his home August 2, 1950. He was graduated from the University and Bellevue Hospital Medical College, New York, in 1904. At the time of his death, he was the Consulting Orthopaedic Surgeon at the Hartford Hospital and the Institute of Living (Neuropsychiatric Institute of the Hartford Retreat), Hartford, the Charlotte Hungerford Hospital in Torrington, the Newington Home for Crippled Children in Newington, the Litchfield County Hospital in Winsted, the Manchester Memorial Hospital in Manchester, the Bristol Hospital in Bristol, the Windham Community Memorial Hospital in Willimantic, and the William W. Backus Hospital, Norwich; he was consulting surgeon of the New Britain General Hospital in New Britain, and the Cape Cod Hospital, Hyannis, Massachusetts. He had been certified by The American Board of Orthopaedic Surgery; he had served as Vice-President of the Connecticut Society for Mental Hygiene; he was a member of The American Orthopaedic Association, and the New England Surgical Society; and he was a fellow of The American Academy of Orthopaedic Surgeons and of the American College of Surgeons.

After a forced early retirement about ten years ago, he acquired a facility in writing, so that his contributions to journals were masterpieces of simplicity and expression. Through research for his editorials and anonymous exchange columns in the Connecticut State Medical Journal, he acquired such a knowledge of medical literature that he was one of the best informed men of our time. Although retirement was not easy for him, he lived it so well that he became a pattern for many men in his community.

For many years Dr. Swett was interested in dairy farming and insisted on almost surgical cleanliness in his barns, animals, and farmers. He believed that raw, certified milk was superior to the dirty but pasteurized product. In his opinion, milk was for babies and not for adults, and he believed that many of the ills of our civilization and the development of allergies could be traced to this source.

Dr. Swett was a devoted family man, and his interests centered there rather than in games and sports. He was possessed of a charming personality and readily made friends. His presence at a medical meeting distinctly added to the pleasure of those attending. As a presiding officer, he acted with dignity and assurance.

Paul Swett will be mourned by his friends and associates. The State of Connecticut and the medical profession will feel keenly the loss of his contributions and of his excellent judgment as a consultant.

JAMES W. TOUMEY, JR.
1900–1950

"Jim" Toumey died in the New England Baptist Hospital on October 18, 1950, after a progressive illness. During this illness, he exhibited one of the most outstanding examples of courage and cheerfulness that the attending staff has witnessed.

Born September 14, 1900, in New Haven, Connecticut, Dr. Toumey received the degrees of B.A. from Yale University in 1922, and of M.D. from the College of Physicians and Surgeons, Columbia University, in 1926. Then followed a surgical internship at the Presbyterian Hospital in the City of New York, three years of training as an intern at the New York Orthopaedic Dispensary and Hospital, and thereafter a fellowship at this same institution for which, in 1933, he received the degree of D.M.Sc. in Orthopaedic Surgery from Columbia University. From 1933 to 1938 he was on duty at this Hospital as a staff surgeon.

In 1938, Jim became a member of the Department of Orthopaedic Surgery at the Lahey Clinic. He continued on that Service until his death, except for an absence while on active duty with the Navy from March 1942 to December 1945, when he assumed an inactive-duty status with the rank of Commander.

Dr. Toumey was an orthopaedic surgeon on the Staffs of the New England Baptist and the New England Deaconess Hospitals and a consultant in the same field at the United States Naval Hospital, Chelsea, Massachusetts. He was a member of The American Orthopaedic Association, the Interurban Orthopedic Club, and the American Medical Association; he was a fellow of The American Academy of Orthopaedic Surgeons, and of the American College of Surgeons.

His contributions to the medical literature covered a wide field, beginning with his initial paper, published in 1934, on "Hip-Joint Tuberculosis Treated by Fusion Operation," through such problems as are presented by intervertebral-disc protrusion, surgery of arthritis of the hip and knee, an effective technique for spine fusion, and the management of metastatic malignant lesions of the spine. The papers more recently read, particularly his paper on reflex sympathetic dystrophy, and his last paper on cauda equina tumors as a cause of the low-back syndrome, will be well remembered by the present members of the societies.

He was a skilled surgeon and was possessed of an alert mind together with a delightful sense of humor and a never-failing desire to do his best. He is sorely missed by his many friends. He is survived by his wife Noel, a daughter Elenor, and a son James.

Book Reviews

AN ATLAS OF AMPUTATIONS. Donald B. Slocum, M.D., M.S. St. Louis, C. V. Mosby Company, 1949. $20.00.

As Chief of the Amputation Section at Walter Reed General Hospital during World War II, Dr. Slocum is eminently qualified to write on the subject of amputations. His *Atlas* is, however, not based entirely upon the results of recent experience, but presents as well those earlier methods which have proved their worth. Although amputation surgery is one of the oldest of the surgical specialties, most of the refinements of technique have evolved as a result of recent wars, until now the work "is a phase of reconstruction employing plastic and orthopedic surgery in delicate balance".

The *Atlas* is divided into four parts. First is given an orientation which includes consideration of the indications for amputation and the ultimate objectives. The second part deals with surgical considerations, including wound healing, care of tissues, surgical preparation, and anaesthesia. Emphasis is placed upon the quality of the stump and its ability to retain a useful prosthesis.

Part 3 contains a discussion of surgical techniques for both open and closed amputations. Here the author has presented and evaluated operative methods, always with a view to their practical significance. Amputations and disarticulations at various levels are described. The operative procedures are illustrated step by step, with photographs at progressive stages of healing.

Part 4, "The Convalescent Period", comprises approximately half of the volume, in keeping with Dr. Slocum's emphasis upon the fact that amputation surgery is not limited to the operative wound, but includes proper fitting of a prosthesis and successful rehabilitation of the amputee to a useful position in society. Of especial interest is an analysis of the normal and amputee gait. In addition, prostheses are shown for various types of amputations.

This book has been very well prepared and represents a complete, up-to-date, authoritative source of information for all phases of amputation therapy. The illustrations are profuse and of excellent quality. Dr. Slocum is to be congratulated on an exceptionally fine piece of work.

REGIONAL ORTHOPAEDIC SURGERY. Paul C. Colonna, M.D. Philadelphia, W. B. Saunders Co., 1950. $11.50.

This textbook of 700 pages, as Dr. Colonna points out in the preface, is a reflection of personal experience as a teacher of graduates and undergraduates. Using the regional approach and stressing diagnosis and treatment, the author has admirably succeeded in covering the subject, including fractures, in a text of reasonable dimensions. Where so much ground has been covered, one might hardly expect to find sufficient essential information given on any one subject. Yet this the author does for the most part, and does well.

The introductory chapters on physiology and general pathology of bones and joints and on orthopaedic examination are excellent and form a sound basis upon which have been built the succeeding chapters, each devoted to a major region of the body.

Special chapters have been added on neuromuscular disabilities, physical medicine (by Dr. George Morris Piersol), and, especially well done, a chapter on tumors of bone.

The illustrations are excellent. The photographs, roentgenograms, and line drawings have been selected carefully and with the instinct of the born teacher to make the idea vivid to the student.

The general make-up of the book is first class and reflects credit on the publishers.

The book is dedicated to Dr. Royal Whitman, whose influence as an inspiring teacher and fearless pioneer may be felt throughout its pages.

Dr. Colonna's broad experience and undoubted love of teaching has made it possible for him to contribute a textbook of real merit and value.

UNFALLCHIRURGIE IM RÖNTGENBILDE. Dr. Walther Ehalt. Wien, Wilhelm Maudrich, 1950.

This book represents an interesting approach to an important problem,—the roentgenographic aspects of traumatic medicine. It should be of value not only to the orthopaedic surgeon, but also to the many physicians in general practice who deal with accident patients in their offices, in insurance work, and in court. Its primary contribution lies in the simple and well recognized fact that today the handling of accident work without the proper use of roentgenographic studies and consultations is impossible and dangerous.

The book is divided into two parts. Part I deals with the roentgenographic approach to the general problems of accurate and complete examination, including technique, evaluation of the findings, treatment and healing, and the recognition of subsequent complications. In Part II, these problems are correlated to the various parts and the specific bones and joints of the body.

The physical background of the roentgenographic examination is described briefly but precisely, and in sufficient detail for a book of this type. Various methods of examination and technical factors essential for a complete study of an accident case are discussed. Stress is laid upon correct positioning and proper identification and dating of films, with particular emphasis on the legal consequences which may arise from neglect of such simple matters as these. The necessity for a two-plane examination, as well as the indications for stereoscopic and comparative films, is explained. Ehalt emphasizes and illustrates the importance of recognizing developmental variations, particularly in growing bone.

Various modes of injury and their recognition by means of adequate roentgenographic examination are discussed. The shape and type of fractures, torsion, bending, or comminution of the bone,—all may be significant. The differential diagnosis of old and recent fractures is explained. Emphasis is placed upon the difficulty of interpretation when joints are involved by fractures extending into them or by partial or complete dislocation. Stress fractures, spontaneous fractures, and pathological fractures are dealt with in separate chapters.

Careful, detailed roentgenographic examination should never be neglected even in those cases in which on superficial observation the injury appears to be slight. Serious changes, such as the dislocation of the carpal lunate or fractures of the calcaneus, the base of the skull, or vertebral bodies, may be detected by roentgenographic studies.

The formation of callus and complications of healing, such as pseudarthrosis and decalcification, are discussed; other chapters take up the problems of traumatic arthritis, aseptic necrosis, and osteochondritis.

The treatment and prognosis in fractures and other forms of injury are covered. Mention is made of soft-tissue changes, — such as myositis ossificans, periosteal calcification, calcification in bursae, tendon, and tendon sheath, and infection.

The excellent quality of the paper and the print makes for easy reading. The text is illustrated with numerous roentgenographic reproductions, demonstrating the points which the author wished to impress upon his reader. There is a brief but satisfactory bibliography and an adequate index.

The author, a student of Dr. Lorenz Böhler, whose pioneer work in many phases of traumatic surgery is well known in this country, speaks with authority. The material on which his book is based embraces over a million roentgenograms. It is recommended most highly to those whose work brings them in contact with this increasingly important phase of medical practice.

INSTRUCTIONAL COURSE LECTURES. THE AMERICAN ACADEMY OF ORTHOPAEDIC SURGEONS. Volume VII. Edited by Charles N. Pease, M.D., and Sam W. Banks, M.D. Ann Arbor, Michigan, J. W. Edwards Co., 1950. $6.50.

The importance of the Instructional Course Lectures given in connection with the Annual Meeting of The American Academy of Orthopaedic Surgeons is recognized. The publication of the lectures given at these sessions makes this valuable material available to many who cannot attend the courses.

The 1950 Volume includes many important articles and they are well presented. Many of these lectures, as published, include comprehensive bibliographies which make them of greater value. The Volume is well indexed. Even the authors of articles listed in these bibliographies are included in the Author Index.

The tables and charts are well presented, as well as the line cuts and some of the drawings; but the wisdom of including roentgenograms and photomicrographs in a book to be lithoprinted may be questioned. Many of these reproductions seem not clear enough to justify the use of space.

The editors must have devoted a great deal of time to the preparation of this volume and they are to be congratulated upon giving to the profession such a valuable work.

SURGERY OF THE SHOULDER. A. F. DePalma, M.D. Philadelphia, J. B. Lippincott Co., 1950. $17.50.

In this source book of only 438 pages is much that is essential of our present knowledge of the anatomy, physiology, and pathology of the shoulder. Of special interest and importance are the chapters which present the author's extensive anatomical material, demonstrating normal variations in structure and the degenerative changes within the scapulohumeral joint, which progress with each of the later decades.

The various disabilities of the shoulder and their management are considered in detail, including chapters on rupture of the musculotendinous cuff, the so-called frozen shoulder, calcareous deposit in the rotator tendons, dislocation, fracture, shoulder pain of neurogenic origin and obstetrical paralysis, bone tumors, and surgical approaches and procedures.

The pages are profusely and handsomely illustrated by photographs, half-tones, and line drawings; the roentgenograms and photomicrographs are exceptionally clear.

As an expression of American medical publishing, this is an outstanding piece of work.

Dr. DePalma has compiled a reference book. But it is more than that, for his book contains much that is, in fact, an original contribution.

FRACTURES AND DISLOCATIONS IN GENERAL PRACTICE. Ed. 2. John Hosford, M.S. (Lond.), F.R.C.S. (Revised by W. D. Coltart, M.B., B.S. (Lond.), F.R.C.S. London, H. K. Lewis, 21 shillings; New York, Paul B. Hoeber, Inc., 1950. $5.00.

Upon first glance at this small textbook, one gains the impression that it is of relatively little value. After reading it through, however, one is convinced that it is a very worth-while text, particularly for the undergraduate and for the practitioner or the general surgeon who handles few fractures and is only moderately experienced in the management of such injuries. It is not profusely illustrated, but the figures which are used are well chosen; the text is extremely well written, can be easily read, and reveals the fact that the knowledge of the authors is considerable. They have presented not only methods of management of individual fractures and dislocations, but also several chapters on fundamental principles of bone healing and basic concepts of fracture management.

Not all authorities would agree, of course, on every method of treatment recommended. This is true, however, of any textbook. The remarks on methods of taking roentgenographic films and the importance of active physical therapy carried out by a cooperative patient, as well as paragraphs on the use of various types of anaesthesia and a chapter on complications of fracture treatment, are some of the highlights.

It was somewhat surprising to see that the authors did not advocate the use of the oscillating motor saw for the removal of plaster casings, and did advocate the application of powdered penicillin or the sulfonamides in fresh wounds, as well as prolonged traction for forearm fractures, and the use of the Whitman plaster spica for management of some hip fractures. These are a few recommendations to which a surgeon experienced in the management of fractures and dislocations would take exception.

While this book by Hosford and Coltart is another of the small texts on fractures and dislocations, it is probably the best written and most easily read book that the writer has seen.

ON THE CONTRIBUTIONS OF HUGH OWEN THOMAS OF LIVERPOOL, SIR ROBERT JONES OF LIVERPOOL AND LONDON, JOHN RIDLON, M.D., OF NEW YORK AND CHICAGO TO MODERN ORTHOPEDIC SURGERY. H. Winnett Orr, M.D. Springfield, Illinois, Charles C. Thomas, 1949. $4.50.

In this book Dr. Orr pays tribute to Hugh Owen Thomas and Sir Robert Jones for their teachings in the field of orthopaedic surgery. He feels that time has shown the soundness of the principles for which they stood. He states:

> "Modern orthopedic surgery has been influenced to a considerable degree by the splints and the fundamental teachings of Hugh Owen Thomas. But Thomas' influence has been much less than it should have been for these reasons:
>
> "1. Thomas' writings were not easily read or understood. They were never indexed, and they are long out of print—but they are classics or even masterpieces of orthopedic surgery.
>
> "2. Thomas' teachings, while they have provided us, in the words of Sir Robert Jones, with an 'orthopedic conscience', have been imperfectly preserved and transmitted to us even by his most loyal pupils—Dr. John Ridlon and Sir Robert Jones.
>
> "3. The fundamental factor of rest as a therapeutic agent has never been understood or applied by anyone . . . as well as by Hugh Owen Thomas and . . . no one else has been as judicious as to the employment of operative surgery, when necessary, or the resumption of function upon recovery, as Thomas."

To prove his statements, Dr. Orr has culled many choice bits from Thomas's little known writings and incidents from his interesting life; these constitute the largest portion of the text. Handicapped by poor health, the fact that his formal education was obtained under the greatest difficulties, and the prejudice which prevented this son of a bone-setter from being accepted readily in professional circles, Thomas became successful because of his adherence to these principles and his devotion to his patients.

All who visited him in his office, at 11 Nelson Street, were impressed by his tireless energy, his long hours of devoted service, and the originality of his methods.

In this office, he was joined by his nephew, Robert Jones; they continued in practice at this address for forty-seven years. A man of warm friendships, pleasing personality, and recognized ability, Robert Jones was given hospital appointments, teaching opportunities, and offices in local, national, and international societies, which were never accorded to Thomas. His conspicuous contributions to the surgery of World War I, as well as his hospital at Baschurch and the orthopaedic centers which he developed during the War won for him national and international fame. "His enormous industry, as evidenced by the large number of operations done by him throughout his career, as well as his extraordinary skill and technical ability, were recognized by surgeons from all over the world, who visited him at Liverpool, and who invited him to their hospitals and to operate in their clinics."

Dr. John Ridlon was one of the first American surgeons to recognize the true value of the work of Hugh Owen Thomas. He returned from his trips to England determined to extend the influence of this man, and in all of his teachings he emphasized the principles which were fundamental to the work of Thomas. His insistence upon adherence to these principles cost him his position at St. Luke's Hospital in New York;

but, as Professor of Orthopaedic Surgery at Northwestern University in Chicago, at other medical schools in Chicago, and on the staffs of various hospitals, he had ample opportunity during his long life to extoll the teachings of Hugh Owen Thomas, as well as the work of Sir Robert Jones. More than any of his contemporaries, he appreciated the points of view of these two men.

One incident which Dr. Orr recalls is a statement often made by Ridlon, that Thomas himself gave him assurance that he counted upon him to carry on, in the United States, the teachings which he had expounded in Great Britain. Thomas considered the establishment of these fundamental principles to be of first importance in the correct treatment of diseases of the bones and joints. He recognized in Ridlon the determination and devotion to a cause which made him feel it his duty to take a stand for what he considered to be right, regardless of the cost.

Dr. Orr feels that his own method of treatment of osteomyelitis by protection of wounds against chemical irritation and secondary infection (through infrequent dressings), and better control of these wounds and fractures by skeletal fixation in plaster-of-Paris casts, is essentially an extension of Thomas's teachings.

Dr. Orr gives generous acknowledgement to Dr. Ridlon for the part which he played in the development of orthopaedic surgery in the United States. For many years he dominated The American Orthopaedic Association. He served as Secretary for sixteen years, and at that time the Secretary was also Editor of the *Transactions*, which preceded *The Journal*. During his years of teaching in Chicago, Dr. Ridlon numbered among his students many of the men who became leaders in orthopaedic surgery in the West during the generation which followed.

The book closes with a beautiful tribute, "Ridlon and His Share in Molding Orthopedic Surgery", by Arthur Steindler, who appreciated the fine qualities of this man. Dr. Steindler, as one of his devoted pupils, rightly accords him a place as one of the great leaders in American orthopaedic surgery. Dr. Steindler writes:

> ". . . for an all-round mastery of the clinical situation, Ridlon had few equals in his field. He combined the soundness and practical sense of H. O. Thomas with the vision and gentleness of Robert Jones. We must recognize in him his straightforwardness, sometimes bluntness, because it stemmed from an inherent passion for truth which, as with Thomas, tolerated no compromise."

A valuable bibliography of articles and books of historical interest is included.

INTER-ALLIED CONFERENCES ON WAR MEDICINE, 1942–1945. Convened by the Royal Society of Medicine. Honorary Editor: Major-General Sir Henry Letheby Tidy, K.B.E., M.D. London, Staples Press Limited, 1947.

On December 7, 1942, the first meeting of the Inter-Allied Conferences on War Medicine was held in London; the last meeting took place in July of 1945. These conferences were held throughout the War and touch upon nearly every phase of medicine as it related to the planning, fighting, and winning of the War. Problems of the different battle fronts are described, in reports written when these problems were actually being met and solved. The value of this general reporting and pooling of medical experience during the fighting of the War must have been invaluable in the betterment of care of military personnel.

The subjects covered in this volume are wide and varied, and they were presented to the Conferences by representatives of various countries. They have not been uniformly edited, so that each chapter has a newness and freshness and a touch of the personality of the man who prepared it.

In addition to the general accounts of the various campaigns, there are various special subjects such as personal experiences, a discussion of German concentration camps, and a fascinating description of D-Day from the medical point of view.

ACCESSORY BONES OF THE HUMAN FOOT. A RADIOLOGICAL, HISTO-EMBRYOLOGICAL, COMPARATIVE-ANATOMICAL, AND GENETIC STUDY. Dyre Trolle. Copenhagen, Einar Munksgaard, 1948.

The highly specialized nature of this monograph may limit its appeal to those engaged in research and study, but it has obviously been prepared with great care and attention, and the translation from the Danish lacks a great deal of the awkwardness which so many scientific translations show. The writer presents the results of numerous tests of earlier histo-embryological investigations which he carried out in the course of long and involved research to determine if bones preformed in hyaline cartilage "do exist at all". The evolution of the primitive foot is discussed, and the possibilities of the origin of the accessory bones are taken up in detail.

The writer's conclusions point to the following possibilities: (1) The existence of an independent hyaline-cartilaginous primordium, of phylogenetic, neogenetic, or anomalous origin; (2) a process of coalescence for an inconstant ossification center, which may be derived either from an originally independent element preformed in hyaline cartilage or from a secondary ossification center of unknown origin; (3) a tendon bone, such as a postnatal formation developed on account of local irritants acting from without; and (4) a non-recognized pathological lesion.

There is a summary in Danish, and a bibliography of over 400 references.